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# Getting a grip on hand use in unilateral cerebral palsy



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# Getting a grip on hand use in unilateral cerebral palsy

## Proefschrift

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# 1

## General Introduction



The ease with which we perform our daily activities makes us almost unaware of how we use our hands. Although we have a dominant hand, in many bimanual activities it is difficult to recognize which hand has a leading role and which hand is assisting as both our hands have more or less equal abilities and can quickly be used alternately to grasp, hold and release. It is when we are mastering a new fine motor activity or during a task that requires refined movements and precision that hand roles become more prominent. In children with unilateral cerebral palsy there is often a discrepancy in the ability of the two hands, and the assisting role of the affected hand is limited. As a consequence of the impairments, bimanual activities may be hampered, which often restricts these unilaterally affected children to fully participate in daily life activities.

The studies described in this thesis investigate the effectiveness of new upper limb interventions for children with unilateral cerebral palsy. These interventions aim to improve the assisting role of the affected hand in bimanual activities and to achieve individually set functional goals. Additionally, in order to improve and refine the assessment of the affected hand and its contribution to bimanual tasks, existing instruments are revised and a new questionnaire is developed. The clinimetric properties of these instruments are scrutinized.

## Unilateral Cerebral Palsy

Cerebral palsy (CP) is the umbrella term for a group of permanent disorders of the development of movement and posture attributable to non-progressive disturbances that occurred in the developing fetal or infant brain.<sup>1</sup> With a prevalence of 2 to 2.5 per 1000 living births, CP is one of the most common causes of motor impairments in childhood.<sup>2,3</sup> About 30% of this population can be classified as unilateral spastic CP, based on neurological subtypes and anatomic distribution of motor impairments. Motor impairments are mostly characterized by increased muscle tone and muscle weakness. In children with unilateral CP, the upper limb is typically more affected than the lower limb. Gross motor abilities in this group are mostly classified as 'walking independently' (i.e., levels 1 and 2 of the Gross Motor Functional Classification System (GMFCS))<sup>4,5</sup> (see appendix). Their manual ability is generally classified as 'with limitations but mostly independent' (i.e., levels 1 and 2 of the Manual Ability Classification System (MACS))<sup>6,7</sup> (see appendix). However, the diversity of the impact of the upper limb deficits becomes apparent in the broad range in which the *functional* use of the affected hand is classified. The House functional classification<sup>8</sup> (see appendix) usually ranges between 'a fair passive assisting hand' (level 2) and a hand that 'easily performs its part in bimanual activities' (level 7) in 95% of the children with unilateral CP.<sup>6,9</sup> Still, not the motor impairments per se affect the ability to move the arm and hand in a smooth and coordinated manner with sufficient variation and range of motion. Rather, actual hand use is affected by a diversity of other

factors related to the brain lesion<sup>1</sup> that influence motor planning and the performance of bimanual activities. Motor planning, i.e. the ability to plan and prepare a motor action before it is executed, may be compromised in children with unilateral CP and can negatively affect the performance of activities beyond problems with movement execution.<sup>10-12</sup> It is suggested that planning problems are due to the inability to integrate available sensory information with the motor commands to the affected hand<sup>13,14</sup>. Sensory deficits are common in unilateral CP.<sup>15, 16</sup> Lack of sensory information and impaired sensorimotor integration influence feedback mechanisms and impact negatively on precision grip and anticipatory control.<sup>17</sup> Sensory deficits impair grasping and manipulation of objects, as a result of which children need to rely more on visual feedback. Increased visual dependency requires extra attentional resources and slows down the performance of bimanual activities. Another factor is the occurrence of mirror movements. These are frequently reported in unilateral CP and have a negative impact on the performance of bimanual activities, regardless of the unimanual capacities.<sup>18</sup> Mirror movements are typically observed on the not-affected side and probably reflect the compensatory recruitment of contralesional brain areas when the primary intention is to move the affected hand.<sup>19, 20</sup> It is postulated that the occurrence of mirror movements might explain that bimanual tasks are often executed without the use of the affected hand and other body parts are used for holding or stabilizing objects.<sup>18</sup> Hence, using the affected hand as an assisting hand in bimanual activities may be challenging for children with unilateral CP and may result in limitations in the independent execution of daily life activities. This in turn may lead to limited societal participation and reduced quality of life. Skold et al<sup>21</sup> emphasized the influence of external factors in the physical and social environment on motor performance. Their study described how children were struggling to choose the most adequate strategy to perform activities. Children frequently decided not to perform an activity after estimating the feasibility of a successful performance and considering the impression their performance would make on their peers. This implies that treatment programs for children with unilateral CP not only need to focus on improving upper-limb capacities for optimal use of the affected arm and hand in day-to-day bimanual activities, but also need to put emphasis on individual needs for strategies to achieve an acceptable level of functional independence at home and in society.<sup>22</sup>

## **Capacity, performance and developmental disregard**

The concepts of 'capacity' and 'performance' in relation to activities were launched with the publication of the International Classification of Functioning, Disability and Health (ICF) in 2001.<sup>23</sup> The ICF defines 'activity' as a task or action executed by a person and introduces 'capacity' and 'performance' as qualifiers of the activity level. 'Capacity' concerns the execution of a task in a controlled environment, such as during a test or

training (i.e., what is a child capable of in a structured environment) and refers to the highest possible level of functioning. 'Performance' refers to the execution of a task in a natural environment. It concerns what is actually done in daily life, influenced by environmental and personal factors. For example, a child might have the capacity to close the zipper of his jacket independently during a therapy session, but at home he closes his jacket with the help from his parents, while at school he immediately asks help from the teacher to be able to quickly go outside and play with his peers. Although a strong relationship was found between outcomes for unilateral capacity and bimanual performance,<sup>24</sup> this does not mean that improvements in unimanual capacities automatically lead to improved performance of daily activities.<sup>24,25</sup> Indeed, there is a large group of children that is using their affected hand less frequently than expected based on their capacities.<sup>26-28</sup> This lack of spontaneous use is referred to as developmental disregard (DD). DD can be defined as a failure to use the potential motor functions and capacities of the affected arm and hand in daily life<sup>29</sup>. Several underlying mechanisms of developmental disregard have been proposed. The behavioral reinforcement theory<sup>30</sup> explains DD as the result of consistent negative feedback experienced when the affected upper limb is used without success. From a neuropsychological perspective it has been hypothesized that the attentional demands associated with motor learning limit spontaneous use of the affected hand if a task is not sufficiently automated.<sup>29</sup> In children with unilateral CP DD has also been explained as a neurological phenomenon comparable to post-stroke hemi neglect.<sup>31</sup> From this perspective, DD is associated with a failure to develop or activate neural pathways due to a lack of experience of sensorimotor stimuli during important developmental periods. This lack of experience might lead to an impaired development of skilled movement patterns of the affected upper limb.<sup>32</sup> DD results in ineffective and time-consuming execution of bimanual activities, since the affected hand is not used when needed and compensatory strategies are applied to compensate for the lack of an assisting hand.

## Upper limb training

To improve the skillful use of the affected upper limb and to diminish DD and increase spontaneous hand use specific therapeutic approaches have been developed for children with unilateral CP in the past two decades. The two main motor-learning based approaches are Constraint Induced Movement Therapy (CIMT) and Bimanual Training (BiT). Both treatments incorporate massed practice of the affected upper limb. In addition, CIMT includes restraining of the unaffected upper limb (using a sling or splint) for a certain amount of time during which the affected arm and hand are trained intensively in unimanual activities, whereas in BiT the affected arm and hand are trained in bimanual activities while no external restraint is applied. In CIMT the primary focus is on improving

unilateral movements of the affected arm and hand, however, improvements in upper limb capacity are not automatically generalized into improved performance during daily life. This is especially the case in children with unilateral CP; they never experienced typical bimanual functioning before. Contrary to adults with stroke, they have to develop a motor plan to execute these activities bimanually, using their newly acquired capacities. The combination of CIMT with BiT might increase the effectiveness of the intervention by enhancing the integration of improved upper limb function in bimanual activities. However, the optimal combination and dose of CIMT and/or BiT protocols has not yet been established. There is a large variation in dose and intensity of training (40-120 training hours in 2-10 weeks),<sup>33</sup> in the applied types of restraints and in the context of training (group vs. individual training, clinical vs. home-based setting).

### **Requirements for pediatric CIMT-BiT interventions**

Therapeutic interventions need to be tailored to the age and individual needs of the participating children. Hence, an intervention developed for adults with stroke (i.e., CIMT) needs to be adapted for use in children and adolescents. Thus, the key features of CIMT (i.e., constraint of the non-affected upper limb, massed repetitive practice, and shaping of complex movement patterns of the affected upper limb) need to be modified and applied in a playful and child-friendly training.

For children and their parents the ultimate goal of an intervention is an improved ability to independently perform age-appropriate tasks in daily life (e.g. self-care, play, sports). Goal-directed training promotes functional performance<sup>34</sup> and increases a child's engagement in meaningful activities. The use of personal goals enhances motivation for training and optimizes performance of activities.<sup>35, 36</sup> As a consequence, goal-setting for a pediatric intervention should preferably be done by the participating children. The process of setting and prioritizing attainable goals is, however, difficult and abstract for young children.<sup>37</sup> If a child cannot be engaged in the goal-setting process, parents and therapists need to assure that the selected goals incorporate the child's preferences and interests instead of the goals only being based on the parents' concerns.

Taub et al.<sup>30, 38, 39</sup> have strongly suggested to use a 'transfer package' (i.e., a set of behavioral techniques to facilitate transfer of therapeutic gains from the treatment setting to daily life) as an inextricable part of CIMT interventions in both children and adults. This is in line with parents' concerns about the transfer of learned skills from the therapeutical setting to the home and school.<sup>40</sup> In young children this transfer will primarily be guided by their parents. Therefore, their involvement in, and contribution to the intervention is invaluable for a successful implementation in the home environment. Evidently, parental instruction is an important part of pediatric interventions to provide parents with the necessary tools and knowledge to support their child during and after an intervention. However, when children get older, they strive for more independency and many of their daily activities occur outside the immediate surroundings of their parents. Hence, for older

children and adolescents, it would be in the line of their development to learn to self-monitor the (supportive) use of their affected upper limb during daily activities and become less dependent on being prompted by their parents and/or teachers. Behavioral techniques have been used for teaching self-management, self-regulation or self-monitoring of the consequences of a disease in children with chronic disease.<sup>41-43</sup> In all these studies results were promising and generalization to daily-life situations occurred. These self-management techniques include several steps and start with identifying personal goals followed by the use of self-instruction, self-monitoring and registration, and finally self-evaluation and rewarding.<sup>44</sup> The use of self-management strategies in an upper limb intervention might offer the opportunity to teach the participants to monitor, evaluate and thereby improve the bimanual execution of their activities in daily-life situations. At the same time, parents can be instructed to reduce habitual prompting and to stimulate their child in the implementation of self-management skills at home.

## Upper limb assessment

To establish the effect of a pediatric upper limb intervention an assessment needs to consist of a balanced set of reliable tests that have been validated for the study population regarding age and diagnosis. Outcomes at all the main ICF domains that are targeted by the intervention should be incorporated. In addition, both the unilateral capacity to grasp, hold and release in a structured situation and bimanual performance in real daily life situations should be part of the assessment. In the last decade, several assessments, validated for children with CP, have become available addressing upper limb capacity or performance.

### Unilateral capacity

The upper limb interventions specifically focus on the functional capacities of the affected arm and hand in bimanual activities. Besides reaching for or manipulating an object that is being held, the main functional aspects are the ability to grasp, hold and release objects. Frequently used tests that assess unilateral capacity in children with unilateral CP are the Quality of Upper Extremity Skills Test<sup>45</sup> and the Melbourne Assessment of Unilateral Upper Limb Function.<sup>46, 47</sup> However, in both these tests only a few items relate to grasping, holding and releasing an object. The test-items and total score reflect a large range of components contributing to upper limb function and movement (e.g. range of motion, fluency, accuracy, dissociated movements, grasp/release, weight bearing, protective extension), but they foremost evaluate concepts at the ICF level of body functions and structures rather than at the activity level.<sup>48, 49</sup> Other frequently used tests for capacity do focus on grasping, holding and releasing (e.g. Jebsen-Taylor test,<sup>50</sup> Box and Blocks test<sup>51</sup>) but they are testing speed and accuracy. Moreover, they are norm-referenced tests for



adults and typically developing children. As a consequence, they produce less reliable results for young children<sup>52</sup> and can be difficult and frustrating for children with minimal capacities. The Modified House Classification<sup>53</sup> was developed for children with CP and consists of 32 items to assess reaching, grasping, holding, releasing and manipulating a diversity of objects without time constraints. Although developed to support the classification of the affected hand, it was reported that the items-scores can be summed.<sup>53</sup> A sum score (i.e. the number of passed items) represents the functional capacity of the assessed hand. However, in order to use the sum score of a scale, it is necessary that the scale is unidimensional (i.e. all items measure the same construct). Although the MHC was shown to be reproducible,<sup>53</sup> its unidimensionality or item-hierarchy have never been tested. These clinimetric properties need to be assessed to assure that the MHC sum score is a valid measure of functional capacity of the affected hand in children with unilateral CP.

### **Bimanual performance**

Several aspects of performance are addressed in frequently used tests and questionnaires, validated for children with CP. The ABILHAND-Kids questionnaire can be used to assess the manual ability to perform daily activities.<sup>54</sup> The Children's Hand-use Experience Questionnaire assesses whether a child uses one or both hands in bimanual activities and how the child perceives its performance when using the affected hand.<sup>55,56</sup> How effectively the affected hand is used when handling objects that require bimanual hand use is measured with a clinical test: the Assisting Hand Assessment.<sup>57, 58</sup> However, to indicate 'underuse' of the affected arm and hand, valid and reliable measures are needed to establish *how often* the affected hand is used spontaneously while performing bimanual activities. The Video Observation Aarts and Aarts module Determine Developmental Disregard (VOAA-DDD)<sup>59</sup> was developed to establish the amount of spontaneous use of the affected hand in two standardized bimanual activities. One activity (stringing beads) was set up in such way that it demands bimanual task execution, while the other activity (decorating a muffin) merely stimulates the use of the affected hand to perform the activity. Developmental disregard is operationalized by a minimal difference in duration of hand use between both activities. To improve the contrast between 'demanding' and 'stimulating' bimanual task execution, the activities of the VOAA-DDD were adapted and the score system was refined.

A parent-reported measure to specifically establish the amount of hand use during every-day activities may be a valuable addition to the above-mentioned outcome measures. Currently, the only parent-completed measure for real-world amount of use of the affected arm is the Pediatric Motor Activity Log Revised (PMAL-R).<sup>60</sup> It is completed during a 30-minute semi-structured interview and evaluates the quality ('how well') and frequency ('how often') of the affected upper limb use. However, sixteen out of the twenty-two PMAL items are unimanual activities, whereas most daily activities are bimanual.<sup>61</sup> No evidence was published for the item selection process of the PMAL-R and

clinimetric properties of the 'how often' scale were not evaluated, but evidence for test-retest reliability, internal consistency and sensitivity to change has been reported.<sup>60</sup> Although a parent-rated questionnaire may not be completely free from subjectivity, more objective measures like accelerometry cannot yet be used in young children,<sup>62</sup> while other techniques (e.g. standardized home/video observation) are time-consuming and might influence natural behavior. To assess the amount of spontaneous use of the affected hand in the home environment, the chosen activities need to be executed at home, be typical daily life activities of children, and be observable by parents. The bimanual activities should vary in the degree to which they elicit bimanual task approach to capture hand-use in children with high and low amounts of spontaneous hand use. As the affected hand is rarely used to perform unimanual activities, it has to be tested whether unimanual activities can be validly used in a scale for spontaneous hand use.

## Aim and outline of this thesis

Based on the available evidence for intensive and task-specific training of the affected upper limb in children with unilateral CP, we hypothesized that a combination of modified CIMT (mCIMT) and BIT would maximize the functional gains in these children. Two mCIMT-BiT intervention protocols were developed to accommodate the needs of children with unilateral CP within the Dutch healthcare context. In both interventions the training was focused on improving upper limb capacity, bimanual performance and the amount of use of the affected arm and hand. On the one hand, a child-friendly mCIMT-BiT protocol for young children (2.5 - 8 years) was developed (the Pirate group-intervention<sup>63</sup>). This group intervention provides 54 hours of massed unilateral training followed by 18 hours of goal-directed and task-specific bimanual training to enhance motor learning in bimanual activities (i.e. play and self-care). On the other hand, for older children (8-18 years) with unilateral CP, we developed a day-camp intervention that combines mCIMT with BiT condensed in one week during school holidays. Given the 'minimal' dose of training (40 hours) in the latter intervention, it was deemed essential to maximize the training effects in terms of amount of upper limb use by implementing self-management techniques. In this way, we aimed to facilitate the generalization of affected hand use toward daily-life bimanual activities after the intervention. These studies are reported in *part one* of this thesis.

Currently, effects of intensive upper limb interventions can be assessed with valid and reliable outcome measures. Still, there is a need for instruments that can validly and reliably assess the unilateral capacity to handle objects and the amount of spontaneous hand use in natural and daily-life circumstances. From this perspective, we attempted to refine some existing instruments and to develop a new parent-rated questionnaire for use in future upper limb interventions. These studies are reported in *part two* of this thesis.

## Part one

In **chapter 2**, the effectiveness of our 8-week mCIMT-BiT intervention (i.e. the Pirate-group intervention) for children between 2.5 and 8 years is investigated in a randomized controlled trial (RCT). The short-term results of this intervention are compared to those of a control group that receives the same amount of upper limb stimulation. In **chapter 3**, we investigate the progression of manual dexterity and the factors influencing the motor learning curve of the children who participate in the mCIMT-BiT intervention. Additionally, we report of the long-term effectiveness of this intervention. **Chapter 4** reports the results of a feasibility study for a high intensity mCIMT-BiT intervention combined with training of self-management strategies in children aged 8 to 18 years.

## Part two

The study of **chapter 5** investigates the item-hierarchy and validity of the MHC to assess upper limb capacity in children with unilateral CP aged 2 to 18 years. **Chapter 6** focuses on the revision of the VOAA-DDD that had been used to determine the effect of mCIMT-BiT on developmental disregard in the Pirate-group trial (data not published in this thesis). The construct validity, inter-rater, intra-rater and test-retest reliability of the VOAA-DDD-R are investigated in children with unilateral CP. In **chapter 7** a new parent-rated questionnaire (Hand-Use at Home Questionnaire, HUH) is introduced to evaluate the amount of spontaneous hand use in children with unilateral cerebral palsy and neonatal brachial palsy paresis. Both the development of the HUH and its internal structure, unidimensionality and validity are addressed.

Finally, **chapter 8** summarizes the findings of the previous chapters, which is followed by a general discussion and implications for clinical implementation and future research.

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# Appendix

## Classifications for Cerebral Palsy

| GMFCS  | MACS   |
|--|--|
| Abilities and limitations in gross motor function  | Ability to handle objects in daily life  |
| <p>I.</p> <p><b>Walks without Limitations</b></p> <p>Can walk indoors and outdoors and climb stairs without using hands for support. Can perform usual activities such as running and jumping. Has decreased speed, balance and coordination</p>   | <p>I.</p> <p><b>Objects are handled easily and successfully.</b></p> <p>At most, limitations in the ease of performing manual tasks requiring speed and accuracy. However, any limitations in manual abilities do not restrict independence in daily activities</p>  |
| <p>II.</p> <p><b>Walks with Limitations</b></p> <p>Can climb stairs with a railing. Has difficulty with uneven surfaces, inclines or in crowds. Has only minimal ability to run or jump</p>  | <p>II.</p> <p><b>Handles most objects but with somewhat reduced quality and/or speed of achievement.</b></p> <p>Certain activities may be avoided or be achieved with some difficulty; alternative way of performance might be used but manual abilities do not usually restrict independence in daily activities.</p> |
| <p>III.</p> <p><b>Walks Using a Hand-Held Mobility Device</b></p> <p>Walks with assistive mobility devices indoors and outdoors on level surfaces. May be able to climb stairs using a railing. May propel a manual wheelchair and need assistance for long distances or uneven surfaces</p>   | <p>III.</p> <p><b>Handles objects with difficulty – the child will need help to prepare and/or modify activities.</b></p> <p>The performance is slow and achieved with limited success regarding quality and quantity. Activities are performed independently if they have been set up or adapted.</p>                 |
| <p>IV.</p> <p><b>Self-Mobility with Limitations; May Use Powered Mobility</b></p> <p>Walking ability severely limited even with assistive devices. Uses wheelchairs most of the time and may propel own power wheelchair. Standing transfers, with or without assistance.</p>  | <p>IV.</p> <p><b>Handles a limited selection of easily managed objects in adapted situations.</b></p> <p>Performs parts of activities with effort and with limited success. Requires continuous support and assistance and/or adapted equipment, for even partial achievement of the activity</p>                      |
| <p>V.</p> <p><b>Transported in a Wheelchair</b></p> <p>Has physical impairments that restrict voluntary control of movement. Ability to maintain head and neck position against gravity restricted. Impaired in all areas of motor function. Cannot sit or stand independently, even with adaptive equipment. Cannot independently walk but may be able to use powered mobility.</p> | <p>V.</p> <p><b>Does not handle objects and has severely limited ability to perform even simple actions.</b></p> <p>Requires total assistance</p>  |

**HOUSE classification**

Functional use as assisting hand

**8****Spontaneous use complete**Uses hand completely  
independently of other hand**7****Spontaneous use partial**Can perform bimanual activities  
easily and occasionally uses the hand  
spontaneously**6****Good active assist**Can actively grasp object and  
manipulate it as well**5****Fair active assist**Can actively grasp object and  
stabilize it**4****Poor active assist**Can actively grasp object and hold  
it weakly**3****Good passive assist**Can hold object and stabilize it for  
use by other hand**2****Fair passive assist**

Can hold onto object placed in hand

**1****Poor passive assist**

Uses as stabilizing weight only

**0****Does not use**





# Part one

Upper limb interventions in  
unilateral CP



# 2

## Effectiveness of modified constraint-induced movement therapy in children with unilateral spastic cerebral palsy: A randomized controlled trial

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## Abstract

In children with unilateral spastic cerebral palsy (CP), there is only limited evidence for the effectiveness of modified constraint-induced movement therapy (mCIMT). The aim of the study was to investigate whether 6 weeks of mCIMT followed by 2 weeks of bimanual task-specific training (mCIMT-BiT) in children with unilateral spastic CP improves the spontaneous use of the affected limb in both qualitative and quantitative terms more than usual care (UC) of the same duration. Twenty-eight children with unilateral spastic CP with Manual Ability Classification System (MACS) scores I, II, or III and aged 2.5 to 8 years were recruited and randomly allocated to either the mCIMT-BiT group ( $n = 28$ ) (three 3-hour sessions per week: 6 weeks of mCIMT, followed by 2 weeks of task-specific training in goal-directed bimanual play and self-care activities) or to 1.5 hours of more general physical or occupational weekly plus encouragement to use the affected hand for the UC group ( $n = 24$ ). Primary outcome measures were the Assisting Hand Assessment and the ABILHAND-Kids. Secondary outcomes were the Melbourne Assessment of Unilateral Upper Limb Function, the Canadian Occupational Performance Measure, and the Goal Attainment Scale. Except for the Melbourne, all primary and secondary outcome measures demonstrated significant improvements in the mCIMT-BiT group. mCIMT followed by task-specific training of goal-directed bimanual play and self-care activities is an effective intervention to improve the spontaneous use of the more affected upper limb in children with relatively good baseline upper extremity function.

Cerebral palsy (CP) is the most frequent cause of physical disability in children.<sup>1</sup> Particularly in hemiparetic children, the focus of treatment is to improve the functional use of the affected arm and leg. In the rehabilitation of the upper limb, according to the Cochrane review by Hoare et al,<sup>2</sup> therapists working with these children try to encourage movements of the affected arm and hand using either bimanual or repeated unilateral activities. However, intensive verbal and physical prompting is often required to 'force' children to properly execute the training tasks. This need for external stimulation is frustrating for both the child and the therapist and frequently results in unsuccessful and incomplete task execution. Indeed, children with unilateral spastic CP often display a form of learned disuse as in daily life they experience too little incentive to use their affected upper limb during functional tasks, in particular during bilateral activities. Gordon's Hand-Arm Bimanual Intensive Training<sup>3,4</sup> encourages the use of the affected arm and hand, but in children with learned disuse, it is still difficult to achieve adequate bimanual coordination.

Constraint-induced movement therapy (CIMT)<sup>5</sup> is a promising approach for rehabilitation of the upper limb in hemiparetic children with CP. This therapy is based on two principles: (1) constraint of the least affected arm and hand and (2) intensive and frequent training of activities with the affected arm and hand. According to a recent Cochrane review<sup>2</sup> CIMT and modified CIMT (mCIMT) should be distinguished from Forced Use Therapy (FUT), which merely imposes a restraint upon the unaffected upper limb. In the past few years, an increasing number of studies has indicated positive (m)CIMT effects on the potential of the affected arm to assist the unaffected arm during bimanual activities,<sup>6,7</sup> as well as on the quality, speed, and dexterity of upper limb function<sup>7-15</sup>, the spontaneous use of the affected arm,<sup>8,9,11</sup> and the level of independence in self-care.<sup>7-9,12-14</sup> However, the restraint method for the affected arm varied greatly (sling, splint, glove, cast), as well as the intensity and length of the restraint (from 8 hours daily during 2-3 weeks to 2 hours daily for 8 weeks).

Clinical evidence supporting (m)CIMT is limited to 3 controlled trials that found beneficial effects on the speed and dexterity of upper limb movement,<sup>9</sup> on functional use measures,<sup>8,9,12</sup> and on the effectiveness of the assisting hand.<sup>6</sup> However, the strength of evidence from two randomized trials (RCTs)<sup>9,12</sup> was limited due to small sample sizes (22 and 18 subjects,<sup>9,12</sup> respectively) and the fact that the control group received considerably less or even no treatment. As a result, the observed outcomes may have been related to differences in the intensity of therapy rather than the specificity of CIMT. A potential confounding factor in the third study<sup>6</sup> was the non-randomized design in which in the experimental group was recruited from 13 centers for pediatric services in the region of Stockholm, whereas the control group was mainly recruited from other regions in Sweden. Evidence of the efficacy of FUT is limited to 2 RCTs that included 25<sup>14</sup> and 31 children.<sup>13</sup> In both studies, problems with including a comparable control group affected the internal validity. In light of these findings, a Cochrane review<sup>2</sup> concluded that, although these results are encouraging, they are still inconclusive and the effectiveness of (m)CIMT or FUT

should be revealed in future trials to obtain stronger evidence. A further recommendation was that these studies should be appropriately powered and should utilize uniform, objective and valid outcome measures.

Considering the variety in the currently proposed therapeutic (m)CIMT approaches and the limited evidence for their clinical efficacy, it remains a challenge to construct an age-appropriate and appealing program for young children with unilateral spastic cerebral palsy to encourage them to use their affected arm and hand, and to teach them new skills. In addition, such a program should incorporate the parents' needs. To achieve the best results in children with unilateral spastic CP, we assumed that training should (1) be focused on children with unilateral CP, (2) take place in a challenging environment, (3) be sufficiently intensive and given in a relatively short period, (4) preferably take place among peers, (5) focus on meaningful activities, (6) start with unilateral training feasible for young children (mCIMT), and (7) end with goal-directed<sup>16,17</sup> task-specific<sup>18</sup> bimanual training (BiT) to integrate the activated upper limb functions in age-appropriate skills. The objective of this study was, therefore, to investigate whether 6 weeks mCIMT followed by 2 weeks of bimanual training (mCIMT-BiT) in children with unilateral spastic CP improves the spontaneous use of the affected limb in both qualitative and quantitative terms more than usual care (UC) of the same duration.

## Methods

### Participants

Children with unilateral spastic CP were recruited from 8 rehabilitation centers in the Netherlands. The children and their parents were first approached and informed by their treating pediatric physiatrist or occupational therapist. A screening was performed by 2 occupational therapists from the recruiting rehabilitation center. Inclusion criteria were (1) cerebral palsy with a unilateral or severely asymmetric, bilateral spastic movement impairment, (2) age 2.5 to 8 years, and (3) Manual Ability Classification System (MACS)<sup>19</sup> scores I, II or III. Exclusion criteria were (1) intellectual disability such that simple tasks could not be understood or executed (i.e. developmental age below 2 years), (2) inability to combine the study protocol with the regular school program, and (3) inability to walk independently without a walking aid.

### Study design

Thirty-six participants (18 per group) were required to obtain a power of 90% to detect at least a moderate treatment effect (Cohen's  $d$ <sup>20</sup> value > 0.5) on the Assisting Hand Assessment (AHA; standard deviation [SD] = 12.22) and/or ABILHAND-Kids (SD = 5.28) using a 2-sided significance level of 0.05. The SDs were derived from earlier pilot observations in the same target group. Taking into account a maximum attrition rate of

30% (due to the intensity of the program), 52 subjects needed to be randomized. Within 48 hours after inclusion, each participant was randomized to the mCIMT-BiT or UC group by throwing a dice with equal probabilities. Before the start of the intervention period (week 0), all children underwent a comprehensive upper limb evaluation that was repeated at the end of the intervention period (week 9) and again after 8 weeks (follow-up in week 17). At the end of the study protocol (week 17), the children who had been allocated to the UC group were also offered the opportunity to participate in a mCIMT-BiT group. The study was approved by the regional Medical Ethical Committee for Research Involving Human Subjects. Oral and written informed consent was obtained from all parents or caregivers.

**Figure 1** Pirate wearing a sling and waving the sword with the affected arm



## Interventions

### *mCIMT-BiT group*

In the mCIMT- BiT group, also named the 'Pirate group', functional training was given during 3-hour afternoon sessions, 3 days per week for 8 weeks, at the primary rehabilitation center (Sint Maartenskliniek; total therapy time: 9 hours/week). During the first 6 weeks, restraint of the unaffected arm and hand was applied. Children were told that they were pirates and that their best arm was injured and had to be kept in a sling. Their affected arm had to be used for all activities, especially to handle a sword (Figure 1). In all these therapy



sessions the principles of shaping and repetitive task practice<sup>8,21</sup> were applied. Immediate feedback on task performance and results was given. During the last 2 weeks, the emphasis was on task-specific exercises in goal-directed bimanual play and self-care activities without restraint. These 2 weeks were used to train individual goals that were set by the parents, using Goal Attainment Scaling.<sup>22</sup>

Each mCIMT-therapy session during the first 6 weeks started with 30 minutes of group activities including dressing up as a pirate, which was followed by making targeted movements<sup>21</sup> (shoulder abduction, shoulder external rotation, wrist extension) with the sword while singing pirate songs. During the following 60 minutes, individual therapy was given in groups of 6 children by 4 occupational therapists, 1 physical therapist and 1 therapy assistant using shaping and repetitive task practice. Afterwards, the children collectively participated in 30 minutes eating and drinking activities during which the principles of repetitive task practice were maintained as much as possible. After a brief sanitary break, the children participated in activities<sup>21</sup> such as board games, card games, puzzles, arts and crafts during 45 minutes in small groups (2 - 3 children). At the end of each session, during 15 minutes, the children changed into their own clothes and prepared for going home. In addition to these therapy sessions, parents were asked to stimulate their child to use the affected arm and hand at home as much as possible and to register the duration of specific periods of stimulation on the child's daily record form.

### ***UC group***

In the UC group, children received a regular rehabilitation program in one of the participating rehabilitation centers. For 8 weeks, the program included individual occupational therapy (OT) and/or physical therapy (PT) given twice a week in 0.5 - 1 hour sessions (total therapy time: 1.5 hours/week). During each OT or PT session, the child was engaged in exercises aimed to stretch the affected arm, to improve its weight-bearing capacity, and to use the affected arm and hand as a good assist. In addition, parents and teachers were instructed to stimulate the children at least 7.5 hours a week to use the affected arm and hand as an assist in daily activities. Parents and teachers received oral and written instructions about activities they were expected to train at home or at (pre)school. Parents and teachers were asked to register the duration of specific periods of stimulation on the child's daily record form.

Due to the nature of both interventions, it was impossible to blind either subjects or therapists with regard to treatment allocation.

### **Outcome assessments**

The primary outcomes were the AHA and the ABILHAND-Kids. The AHA aims to evaluate the spontaneous use of the assisting hand during activities that require bimanual handling in children in the age range 18 months to 12 years.<sup>23,24</sup> Interrater and intrarater reliability of the AHA<sup>25</sup> have high intraclass correlation coefficients for sum scores (.98 for 2-rater design

**Table 1** Baseline characteristics of both groups<sup>a</sup>

| Characteristics                | mCIMT-BiT<br>(n = 28) | UC<br>(n = 22)  |
|--------------------------------|-----------------------|-----------------|
| Sex                            |                       |                 |
| Male                           | 14 (50)               | 14 (64)         |
| Female                         | 14 (50)               | 8 (36)          |
| Age (mean $\pm$ SD in years)   | 4.8 $\pm$ 1.3         | 5.1 $\pm$ 1.7   |
| Distance (mean $\pm$ SD in km) | 55.0 $\pm$ 35.9       | 45.2 $\pm$ 30.2 |
| 1 – 29                         | 8 (29)                | 8 (36)          |
| 30 – 68                        | 10 (36)               | 8 (36)          |
| 69 – 152                       | 10 (36)               | 6 (27)          |
| Hemi paretic side              |                       |                 |
| Left                           | 14 (50)               | 14 (64)         |
| Right                          | 14 (50)               | 8 (36)          |
| GMFCS                          |                       |                 |
| I                              | 27                    | 21              |
| II                             | 1                     | 1               |
| MACS                           |                       |                 |
| I                              | 9                     | 7               |
| II                             | 12                    | 10              |
| III                            | 7                     | 5               |

<sup>a</sup> Values are the number (percentages) unless otherwise indicated. mCIMT = modified Constraint Induced Movement Therapy Group, UC = Usual Care Group, Distance = number of km between St. Maartenskliniek and home address of the child, GMFCS = Gross Motor Function Classification System, MACS = Manual Ability Classification System.

and .97 for 20-rater design; intrarater = .97). We used scaled scores to compare performances between different weeks. The ABILHAND-Kids is a questionnaire that measures manual skills in children with upper limb impairments.<sup>26,27</sup> This scale consists of 21 mostly bimanual items rated by the parents. Its range and measurement precision are appropriate for clinical practice (reliability:  $R = .94$ ; reproducibility over time:  $R = .91$ ). Whereas the AHA is primarily focused on play activities, the ABILHAND-Kids is an instrument to investigate manual ability in self-care activities.

Secondary outcomes consisted of the Melbourne Assessment of Unilateral Upper Limb Function (Melbourne),<sup>28</sup> the Canadian Occupational Performance Measure (COPM),<sup>29</sup> and Goal Attainment Scaling (GAS).<sup>30</sup> The Melbourne aims to assess the quality of upper limb movements in children with neurological impairment in the age range 5 to 15 years. The modified Melbourne Assessment of Unilateral Upper Limb Function<sup>31</sup> for children in the age range 2 to 5 years has recently been developed. In the present study, this modified Melbourne was used for this age group, whereas the original Melbourne was used for the older children. In both versions of the Melbourne, 16 different tasks are scored from a video tape on range of motion, accuracy and fluency of movement.<sup>32</sup> The COPM is a family-centered tool that lists the problems experienced in daily life through a semi-

structured interview.<sup>29</sup> Five of the most important areas are selected and then scored by the parents at 2 levels: (1) Perception of current performance (COPM-P) and (2) satisfaction with current performance (COPM-S). The COPM-ratings are on a 10-point scale; scores closer to 10 indicate better performance and increased satisfaction. By means of the COPM, individual training goals were set by the parents. With the GAS, the most important functional goal for the affected arm and hand was broken down into attainable sub goals during the pre-intervention assessment with the parents.<sup>30,33</sup> In this study, the perceived outcome was scaled from -3 to +2. A score of -3 indicated a level lower than the initial performance level, -2 indicated an unchanged level of performance, -1 indicated a level lower than the desired outcome, 0 indicated the desired outcome level had been achieved, +1 indicated somewhat more improvement than expected, and +2 indicated much more improvement than expected (in total 6 levels of performance). Parents scored their children at each measurement by selecting the appropriate performance level. All selected outcome measures are valid and reliable, and most of them have been recommended in the literature to evaluate the effects of (m)CIMT.<sup>2,7</sup>

All assessments were conducted by the same occupational therapist at the primary rehabilitation center, who was unaware of the individual study phase of any particular child, blinded for group allocation, and not involved in any other aspect of the study. Both the AHA and the Melbourne tapes were scored by a certified occupational therapist who was blinded for group allocation and test session.

## Statistical analysis

Student *t* tests (for unrelated samples) were used to determine comparability at baseline with regard to socio-demographic characteristics (age, distance between residence and the primary rehabilitation center, as well as all outcome measures except the GAS). For the AHA, ABILHAND-Kids, Melbourne, COPM-P and COPM-S, the 2 groups were compared post treatment (week 9) with ANCOVA in which differences at baseline, even when 'insignificant', were used as covariates. Cohen's *d* values<sup>20</sup> were calculated to obtain a pre-post intervention effect size. The ratings of the GAS were dichotomized into either 'improved' (increase of 2 points or more) or 'not improved' (less than 2 points increase or any decrease); the percentages of children who improved were analyzed by intervention group. To find out if possible effects remained constant over time, Student *t* tests (paired samples) were used to compare the results between week 9 (post intervention) and week 17 (8-week follow up). All data handling and analyses were carried out by an independent statistician who was blinded for group allocation. SPSS version 17.0 was used for computerized analysis (SPSS, Inc., Chicago, IL).

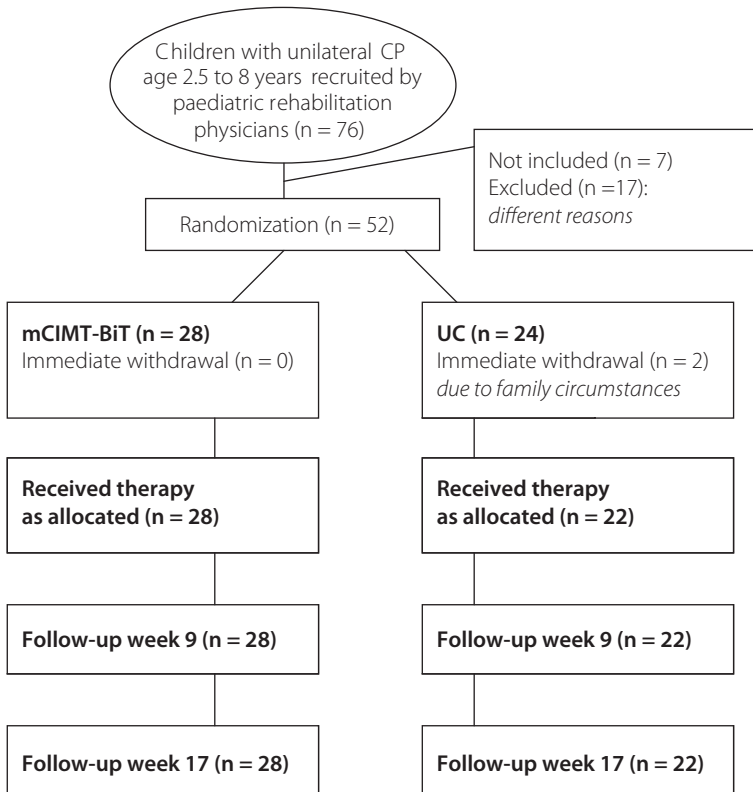
## Results

### Patient flow

Information pertaining to the patient flow through the trial is presented in Figure 2.

In total, 76 children were screened for eligibility. Seven children could not be included because their MACS scores were too high. Seventeen children were excluded: 8 children because of mental retardation, 7 children because they could not combine the training with their school program, and 2 children because they could not walk independently.

**Figure 2** Patient flow diagram



mCIMT-BiT, modified constraint-induced movement therapy combined with bimanual training; UC, usual care.

**Table 2** Primary outcome measures for both groups

| Outcome measures        | mCIMT-BiT   | ΔmCIMT-BiT | UC          | ΔUC       | Mean difference (95% CI) <sup>a</sup> | Effect Size <sup>b</sup> | mCIMT-BiT t-value | mCIMT-BiT p-value | UC t-value | UC p-value |
|-------------------------|-------------|------------|-------------|-----------|---------------------------------------|--------------------------|-------------------|-------------------|------------|------------|
| AHA (range 0 - 100)     |             |            |             |           |                                       |                          |                   |                   |            |            |
| Baseline, mean ± SD     | 53.3 ± 14.6 |            | 50.6 ± 22.5 |           |                                       |                          |                   |                   |            |            |
| Week 9                  | 60.1 ± 15.3 | 6.8 ± 8.2  | 53.1 ± 22.2 | 2.5 ± 6.3 | 4.5 (0.26-8.77)                       | 0.43                     |                   |                   |            |            |
| Week 17                 | 59.7 ± 13.5 | 6.4 ± 5.7  | 52.3 ± 21.4 | 1.7 ± 5.5 |                                       |                          | 0.306             | 0.762             | 0.764      | 0.453      |
| ABILHAND (range 0 - 42) |             |            |             |           |                                       |                          |                   |                   |            |            |
| Baseline, mean ± SD     | 20.9 ± 5.1  |            | 22.6 ± 6.9  |           |                                       |                          |                   |                   |            |            |
| Week 9                  | 28.4 ± 5.9  | 7.5 ± 4.0  | 23.7 ± 6.0  | 1.1 ± 4.8 | 5.9 (3.55-8.29)                       | 1.01                     |                   |                   |            |            |
| Week 17                 | 28.9 ± 5.2  | 8.0 ± 3.9  | 24.4 ± 6.6  | 1.8 ± 3.8 |                                       |                          | -0.817            | 0.421             | -0.816     | 0.424      |

mCIMT-BiT= modified Constraint Induced Movement Therapy combined with Bimanual Training; UC= Usual Care Group; AHA= Assisting Hand Assessment; ABILHAND= ABILHAND-Kids. <sup>a</sup> Mean group difference (95% Confidence Interval) (differences at baseline were used as covariates), <sup>b</sup> Effect Size (Cohen's d): 'small' = 2 < d < 5, 'moderate' = 5 < d < 8, and 'large' = d > 8.

As a result, 52 children with unilateral spastic CP were definitively included. Of these children, 28 children were randomly allocated to the mCIMT-BiT group and 24 to the UC group. Immediately after randomization, 2 children withdrew from the UC group due to family circumstances. Thereafter, no subjects were lost to follow-up or changed group allocation. Hence, the data of 22 subjects in the UC group will be presented.

### Control of bias

The mCIMT-BiT group ( $n = 28$ ) and the UC group ( $n = 22$ ) had similar baseline sociodemographic characteristics (see Table 1). In addition, no significant differences between groups were found for the baseline scores on the AHA, ABILHAND-Kids, Melbourne, COPM-P, or COPM-S. According to the children's daily record forms, the mCIMT-BiT group received on average 9 hours per week therapy and 3.3 hours additional stimulation at home (total stimulation time =  $12.3 \pm 1.9$  hours). The UC group received on average 1.5 hours per week therapy and 11.2 hours additional stimulation at home or at school (total stimulation time =  $12.7 \pm 2.1$  hours). Blinding of the assessor was tested afterwards by asking the OT to indicate for each child whether she thought it had participated in the mCIMT-BiT or in the UC group. Comparable with the actual distribution of the children, the OT assessor made a correct guess in 48% of all cases.

### Primary outcomes

Table 2 presents the results for the primary outcome measures. The mCIMT-BiT group showed significantly more improvement after 9 weeks compared to the UC group.

Compared to baseline (week 0), the children of the mCIMT-BiT group showed a 13% improvement on the AHA as well as a 36% improvement on the ABILHAND-Kids at the end of the intervention (week 9). These improvements were 2.5 and 7 times greater than the respective scores of the UC group: 5% and 5%. The effect size was small for the AHA (Cohen's  $d = .43$ ) and large for the ABILHAND-Kids (Cohen's  $d = 1.01$ ).

At follow-up (week 17), the observed improvements were maintained in the mCIMT-BiT group; compared with week 9 the AHA decreased by 1%, whereas the ABILHAND-Kids increased by 2%. The improvements of the UC group decreased slightly on the AHA by 2% and increased slightly on the ABILHAND-Kids by 3%. Within either group, there were no significant differences between the scores on the AHA and ABILHAND-Kids between week 9 and week 17.

### Secondary outcomes

The results for the secondary outcome measures are presented in Table 3. Overall, the pattern of change was the same as for the primary outcome measures: The mCIMT-BiT group showed significantly greater improvements than the UC group on all secondary outcomes, except for the Melbourne, which merely showed a positive trend in favor of the mCIMT-BiT group. After 9 weeks the mCIMT-BiT group showed improvements on the

**Table 3** Primary outcome measures for both groups

| Outcome measures      | mCIMT-BiT       | ΔmCIMT-BiT | UC              | ΔUC       | Mean difference (95% CI) <sup>a</sup> | Effect Size <sup>b</sup> | mCIMT-BiT t-value | mCIMT-BiT p-value | UC t-value | UC p-value |
|-----------------------|-----------------|------------|-----------------|-----------|---------------------------------------|--------------------------|-------------------|-------------------|------------|------------|
| COPM-S (range 0 - 10) |                 |            |                 |           |                                       |                          |                   |                   |            |            |
| Baseline; mean ± SD   | 3.7 ± 1.1       |            | 3.9 ± 1.1       |           |                                       |                          |                   |                   |            |            |
| Week 9                | 7.4 ± 1.2       | 3.7 ± 1.6  | 5.3 ± 1.2       | 1.4 ± 1.1 | 2.2 (1.51-2.86)                       | 1.32                     |                   |                   |            |            |
| Week 17               | 7.3 ± 1.2       | 3.6 ± 1.6  | 5.5 ± 1.2       | 1.6 ± 1.3 |                                       |                          | 0.288             | 0.776             | -0.949     | 0.535      |
| COPM-P (range 0 - 10) |                 |            |                 |           |                                       |                          |                   |                   |            |            |
| Baseline; mean ± SD   | 3.0 ± 1.0       |            | 3.4 ± 1.1       |           |                                       |                          |                   |                   |            |            |
| Week 9                | 6.5 ± 1.0       | 3.5 ± 1.3  | 4.6 ± 1.4       | 1.2 ± 1.1 | 2.1 (1.43-2.72)                       | 1.31                     |                   |                   |            |            |
| Week 17               | 6.5 ± 0.9       | 3.5 ± 1.3  | 4.7 ± 1.4       | 1.3 ± 1.2 |                                       |                          | -0.503            | 0.619             | -0.527     | 0.604      |
| Melb. (range 0 - 100) |                 |            |                 |           |                                       |                          |                   |                   |            |            |
| Baseline; mean ± SD   | 63.9 ± 12.6     |            | 62.1 ± 16.2     |           |                                       |                          |                   |                   |            |            |
| Week 9                | 68.8 ± 11.6     | 5.0 ± 7.6  | 63.5 ± 16.7     | 1.4 ± 6.2 | 3.8 (-0.06-7.73)                      | 0.40                     |                   |                   |            |            |
| Week 17               | 69.1 ± 12.0     | 5.3 ± 5.8  | 65.1 ± 14.3     | 3.0 ± 6.0 |                                       |                          | -0.318            | 0.753             | -1.410     | 0.173      |
| GAS Goals (%)         |                 |            |                 |           |                                       |                          |                   |                   |            |            |
| Week 9                | 82 <sup>c</sup> |            | 23 <sup>c</sup> |           | 59 <sup>c</sup>                       |                          |                   |                   |            |            |
| Week 17               | 86 <sup>c</sup> |            | 36 <sup>c</sup> |           | 50 <sup>c</sup>                       |                          |                   |                   |            |            |

mCIMT-BiT= modified Constraint Induced Movement Therapy combined with Bimanual Training Group; UC= Usual Care Group; COPM-S= Canadian Occupational Performance Measure-Satisfaction with current performance; COPM-P= Canadian Occupational Performance Measure-Perception of current performance; Melb.= Melbourne Assessment of unilateral upper limb function for children with neurological impairment; GAS= Goal Attainment Scaling (first goal improved with 2 points or more). <sup>a</sup> Mean group difference (95% Confidence Interval) (differences at baseline were used as covariates). <sup>b</sup> Effect Size (Cohen's d): 'small' = .2 < d < .5, 'moderate' = .5 < d < .8, and 'large' = d > .8. <sup>c</sup> Percentage of children that showed an increase of 2 points or more compared to baseline.

COPM-P and the COPM-S with respect to baseline of 117% and 100%, respectively. The UC group showed much lower percentages of change, 35% and 36%, respectively. The effect sizes were large for both the COPM-P and the COPM-S (Cohen's  $d = 1.32$  and  $1.31$ , respectively). Improvements on the Melbourne were 8% and 2% compared to baseline for the mCIMT-BiT group and the UC group, respectively. This group effect was small (Cohen's  $d = 0.4$ ). The dichotomized GAS scores showed improvement (increase of 2 points or more) in 82% of the mCIMT-BiT participants, whereas only 23% of the UC group showed improvement on the GAS.

At follow-up (week 17), the improvements of the mCIMT-BiT group compared to post treatment (week 9) increased by 3% for the COPM-P and decreased by 3% for the COPM-S. The improvements of the UC group increased by 3% and 8% for the COPM-P and the COPM-S, respectively. The improvements on the Melbourne remained stable for the mCIMT-BiT group and increased by 2% for the UC group. As for the GAS, 86% of the mCIMT-BiT and 36% of the UC participants still showed improvement compared with baseline. Within either group, there were no significant differences between the scores on the COPM-P, COPM-S, or the Melbourne between week 9 and week 17.

## Discussion

We investigated whether 8 weeks of mCIMT-BiT in young children with CP improves the spontaneous use of the upper limb in both qualitative and quantitative terms more than UC of the same duration. All primary and secondary outcome measures demonstrated significantly greater improvement in the mCIMT-BiT group than in the control group; the Melbourne showed at most a positive trend in favor of mCIMT-BiT. In comparison with UC, mCIMT-BiT improved the effectiveness of the assisting hand (AHA) and also demonstrated higher scores for bimanual performance during self-care and leisure tasks (the ABILHAND-Kids). All effects were maintained at 8 weeks follow up, which implies a clinically meaningful effect even after the therapy had finished.

A strength of this study is that the Pirate group provided a meaningful and challenging environment for the children, who enjoyed the play-like treatment and the provocative activities with peers. Parents reported that the children liked the therapy in the Pirate group much more than the individual therapy they had received before they started in the Pirate group. In addition, parents were very much engaged in the family-centered process of selecting goals for their child. They often commented that their participation in stimulating the use of the pirate hand at home kept them focused on the intervention and improved the child's compliance with the therapy. In contrast to other studies,<sup>9,11</sup> no child dropped out from the mCIMT-BiT group and none of the children deviated substantially from the study protocol.



The results of this study corroborate the findings of previous studies showing significant improvement in upper limb function through (m)CIMT or FUT in children with CP. However, an essential difference is that in this study improvements were achieved by using a combination of 6 weeks mCIMT and 2 weeks of goal-directed and task-specific bimanual training, each during 3 hours per afternoon and 3 times per week, whereas some other studies have employed much longer periods of physical restraint (6-8 hours per day <sup>4,12</sup>). With 50 participants, this study was appropriately powered, and the randomization procedure created comparable groups at baseline with regard to socio-demographic characteristics and outcome measures. Total stimulation time was comparable between the mCIMT-BiT and the UC groups, and the OT assessor was blinded successfully. The principles of mCIMT-BiT as described by Taub<sup>34</sup> were used and involved 3 main elements: (1) intensive training of the more affected extremity, (2) prolonged restraint of the less affected extremity, and (3) a 'package of techniques' to induce behavioral change in daily life activities.

As suggested by Hoare et al,<sup>2</sup> valid and reliable outcomes that measure the use of the most affected upper limb in bimanual tasks (e.g., the AHA) and its use in relation to individual client and family goals (e.g., the COPM and GAS) were selected. The greatest changes from baseline to post treatment were found for the COPM-P and COPM-S, which is in contrast with the mCIMT feasibility study by Wallen et al,<sup>7</sup> in which only a trend for COPM improvement was found. One explanation for this difference is that this study incorporated 2 weeks of task-specific bimanual training for the mCIMT-BiT group to optimize transfer of upper limb skills to daily life activities. The mCIMT-BiT group showed relatively large improvements on the ABILHAND-Kids as well, which might be related to the same period of 'transfer training'. In this study, all COPM, GAS, and ABILHAND-Kids forms were filled in together with the same OT assessor to overcome the problem mentioned by others that the scoring only by parents may be biased by their subjectivity.<sup>7</sup> Although it showed a positive trend, the improvements of the mCIMT-BiT group on the capacities measured by the Melbourne were small. As in the study by Wallen et al,<sup>7</sup> the changes in the Melbourne did not reach the 12% value to be considered clinically significant. It may well be that the Melbourne is less sensitive to the applied mCIMT-BiT intervention, because it focuses on upper limb capacity rather than spontaneous use of the upper extremity during 'natural' activities. This could also mean that the children might have needed a longer period of restraint and training to reach a significant result on the Melbourne.

Although this is the largest RCT of (m)CIMT in children with CP, the number of participants is still moderate. In addition, most of the children that were included had a relatively good arm-hand capacity, that is, 73.6 % had an MACS score of I or II. Both these aspects limit the generalizability of the results of this study. Another limitation is the possible difference in 'quality of stimulation' between both groups. Although the total duration of therapy plus stimulation was the same, the mCIMT-BiT group was mostly

trained by dedicated therapists, whereas the UC group received therapeutic attention from parents and teachers. Last, the follow-up of 8 weeks was relatively short to judge long-term effects, whereas the loss to follow up of 2 participants in the UC group immediately after randomization prevented a true intention-to-treat analysis.

In conclusion, mCIMT-BiT is an effective intervention to increase the spontaneous use of the more affected arm and hand in children with CP, aged 2.5 to 8 years, with MACS scores I, II or III. Future research might be focused on the preventive effect of mCIMT-BiT in even younger children as well as on the therapeutic effects in children with less arm-hand capacity. In addition, the optimal frequency and intensity of mCIMT-BiT in young children with CP should be determined in relation to the size and duration of its effects, including the need for 'booster' sessions in case of functional relapse.

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# 3

## Motor-learning curve and long-term effectiveness of modified constraint induced movement therapy in children with unilateral cerebral palsy: a randomized controlled trial

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## Abstract

The goal of this study was to determine the progression of manual dexterity during 6 weeks (54 hours) (modified) constraint induced movement therapy ((m)CIMT) followed by 2 weeks (18 hours) bimanual training (BiT) in children with unilateral spastic cerebral palsy (CP), to establish whether and when a maximal training effect was reached and which factors might influence the motor learning curve. In addition, long-term retention of effects was determined. In a randomized controlled trial of 52 children with CP, aged 2.5 to 8 years, comparing mCIMT-BiT to conventional therapy, 28 children were allocated to the mCIMT-BiT group. This group was assessed weekly with the Box and Block test. Long-term effectiveness was determined by collecting follow-up data of the primary (Assisting Hand Assessment, ABILHAND-Kids) and secondary (Melbourne, COPM) outcomes at six months and one year after intervention. Fifteen children (53.6%) reached a maximum training effect within the mCIMT period. This group differed from others with respect to age, but not gender, affected side or manual ability. Children younger than five years had a greater chance to reach a maximum score within 6 weeks mCIMT (OR=6.67, 95%CI=1.24-35.71) that stabilized already after four weeks; older children showed a longer progression and tended to decline afterwards. In both age groups, beneficial effects were retained in the long term. The findings suggest that children of 5 years and older might profit from a longer period of mCIMT than 54 hours to reach their maximum unimanual capacity and to retain this capacity during subsequent bimanual training.

Evidence is cumulating that constraint-induced movement therapy (CIMT) has beneficial effects on the capacity as well as the use of the affected upper limb in children with unilateral spastic Cerebral Palsy (CP).<sup>1-8</sup> In addition, several studies have shown that the beneficial effects of (modified) CIMT / (m)CIMT are retained at six months<sup>2, 3, 6, 8, 9</sup> and even one year<sup>10</sup> post intervention. However, relatively little is known about the learning curve during the period of constraint, for instance, at what point in time (maximal) effects are reached. Systematic reviews on (m)CIMT and forced use therapy have concluded that studies vary widely in terms of intervention characteristics, in particular with regard to the type and overall duration of constraint and the total dosage of unilateral upper-limb training.<sup>11-13</sup> Because mCIMT programs are very intensive and, thus, demanding in terms of time and effort invested by the child and the family, adequate dosing of an mCIMT program is essential to prevent unnecessary burden to the participants and to assure optimal cost-effectiveness. In addition, a well targeted dosage of mCIMT can minimize the potential risk of damage to the immature brain as a consequence of restraining the use of a healthy limb in a young child.<sup>14</sup>

A few studies<sup>2, 15</sup> have been undertaken to find evidence for the best composition of mCIMT, tailored to the needs and capacities of the children. A recent randomized controlled trial (RCT),<sup>2</sup> comparing a six hours/day with a three hours/day protocol (both 21 intervention days) in *young* children (n=18, age 3-6yrs), found no differential effects and suggested that a three hours/day training during four weeks (a total of 63 hrs.) would be sufficient for an optimal effect. On the other hand, a review<sup>15</sup> comparing two mCIMT protocols (60 hrs./10 days<sup>3</sup> vs. 90 hrs./15 days<sup>6</sup>) for *older* children (n=21, age 4-13yrs, and n=20, age 3-10yrs, respectively) showed a larger improvement of hand function as assessed with the Jebsen Taylor test in the group receiving 90 hours of training. Hence, the conclusion is justified that the optimal intensity of mCIMT is still unknown for various age groups with unilateral spastic CP. In addition, it is unknown whether optimal (maximal) effects are being reached at all and which factors (e.g. age, manual ability at baseline, training intensity) might influence the motor learning curve as well as the retention of treatment effects in the long term.

Previously, we have reported on the efficacy of six weeks mCIMT followed by two weeks bilateral training (mCIMT-BiT) in 28 children with unilateral spastic CP, aged 2.5-8 years, providing results up to two months post intervention.<sup>1</sup> Our mCIMT-BiT program was embedded in a playful 'Pirate group' program which facilitated both constraint of the 'wounded' (i.e., non-affected) arm and unilateral training of the 'sound' arm handling the sword (i.e., the affected arm). This program consisted of 54 hours of unilateral upper-limb training followed by 18 hours of bimanual training to ensure that training effects would be integrated into daily life bimanual activities.<sup>16</sup> Although this intervention turned out to be highly effective to improve the use of the affected upper limb as assessed with the Assisting Hand Assessment (AHA)<sup>17</sup> and the ABILHAND-Kids<sup>18</sup>, we could not draw any



conclusions based on these measures with regard to the *learning curve* of the children during the mCIMT period or the BiT period. For this purpose, a different outcome measure was collected every week during the entire training period of eight weeks, i.e., the Box and Block test. This separate measure, focusing on the main functions of the hand (i.e., grasp, hold and release), was selected in order not to influence the assessments of the primary outcomes (AHA, ABILHAND-Kids) due to repeated testing. To the best of our knowledge, there are no previous studies that reported on the learning curve *during* a mCIMT intervention.

Hence, the primary goal of this study was to investigate the progression of manual dexterity as assessed with the Box and Block test during a six-weeks mCIMT program followed by a two-weeks BiT program that has proven its effectiveness in children with unilateral spastic CP, aged 2.5-8 years, in order to determine whether and when a maximum training effect was reached and what factors might influence the learning curve. The ultimate goal was to extract information based on which the optimal dosage of constraint and unilateral upper-limb training could be determined. In addition, we compared the follow-up data of the original primary and secondary outcome measures at six months and one year post intervention with the previously published results one week post intervention for all children that participated in the original study.<sup>1</sup>

## Methods

### Participants and design

Fifty children with unilateral spastic CP, aged 2.5 to 8 years, participated in this study. The selection process has been described previously (Aarts et al., 2010). Table 1 shows the demographic and clinical characteristics of the participants. The Manual Ability Classification System (MACS) was used to classify manual ability, focusing on actual performance of handling objects.<sup>19</sup> Twenty-eight children took part in the mCIMT-BiT group and 22 in the usual care group (UC) after random allocation. Only the 28 subjects participating in the mCIMT-BiT group were tested weekly with the Box and Block test to assess changes in manual dexterity during the intervention. As for the original primary and secondary outcome measures, all children were assessed at baseline and at one week, two months and six months after the intervention.

In addition, the mCIMT-BiT group underwent a follow-up assessment of the primary outcome measures one year after the intervention. In the present study, the follow-up assessments at six months and one year post intervention are compared to the data one week post intervention.

**Table 1** Main characteristics of all participants (n=50)

|                     | mCIMT-BiT group<br>n=28 | UC group<br>n=22 |
|---------------------|-------------------------|------------------|
| Mean age years (SD) | 4.8 (1.3)               | 5.1 (1.7)        |
| Gender              |                         |                  |
| Male                | 14                      | 14               |
| Female              | 14                      | 8                |
| Affected side       |                         |                  |
| Left                | 14                      | 14               |
| Right               | 14                      | 8                |
| MACS                |                         |                  |
| I                   | 9                       | 7                |
| II                  | 12                      | 10               |
| III                 | 7                       | 5                |

mCIMT-BiT= modified Constraint Induced Movement Therapy-Bimanual Training, UC= usual care, MACS=Manual Ability Classification System

## Intervention

The mCIMT-BiT intervention in the Pirate group and the control intervention (usual care of equal intensity and duration) have been described in detail in previous publications.<sup>1, 16, 20</sup> Briefly, the mCIMT-BiT program entailed nine hours training per week (i.e., a total of 72 hours). During the first six weeks unilateral training of the affected ('sound') arm was given during Pirate play activities, while constraint was applied to the non-affected ('wounded') arm by a sling (i.e., 54 hours mCIMT). Thereafter, two weeks of bimanual training were given with emphasis on meaningful daily activities (i.e., 18 hours BiT). Children in the usual care group received a regular rehabilitation program combined with specific stimulation of bimanual hand use at home and in (pre)school, adding up to nine hours a week for eight weeks (i.e., a total of 72 hours). In this group no constraint of the non-affected hand was applied. In both groups, training activities were based on individual goal setting by parents and therapists.

## Outcome measures

### Learning curve

To study the learning curve during the mCIMT-BiT intervention, the Box and Block test was chosen because this measure (1) incorporates the most essential hand functions (i.e., quick coordination of grasp, hold, transfer and release of a small object), (2) is easy to explain and can readily be performed by many children, (3) takes little time to administer (test duration one minute), and (4) shows no ceiling effects. The Box and Block test assesses manual dexterity by counting the number of blocks that are transferred with a single hand from one compartment to another within 60 seconds.<sup>21</sup>

**Table 2** Characteristics of mCIMT-BiT group divided in age groups (n=28)

| Characteristics                                  | <5 years   | ≥ 5 years  | p                  |
|--|------------|------------|--------------------|
| n (%)  | 13 (46,4%) | 15 (53,6%) |                    |
| Mean age<br>years                                | 3.5        | 5.9        |                    |
| Gender   |            |            |                    |
| Male   | 8          | 6          | 0.256 <sup>a</sup> |
| Female   | 5          | 9          |                    |
| Affected side                                    |            |            |                    |
| Left   | 7          | 7          | 0.705 <sup>a</sup> |
| Right  | 6          | 8          |                    |
| MACS   |            |            |                    |
| I  | 5          | 4          | 0.588 <sup>b</sup> |
| II   | 5          | 7          |                    |
| III  | 3          | 4          |                    |
| Top reached within 6 weeks<br>n (%)              | 10 (76,9%) | 5 (33%)    |                    |
| <b>Outcome measures</b>                          |            |            |                    |
| Mean change (baseline - one week post treatment) |            |            |                    |
| AHA (logits)                                     | 1.067      | 1.068      | 0.998 <sup>c</sup> |
| ABILHAND-Kids(logits)                            | 1.205      | 1.360      | 0.628 <sup>c</sup> |
| Melbourne  | 4.15       | 5.71       | 0.599 <sup>c</sup> |
| COPM-P   | 3.692      | 3.387      | 0.549 <sup>c</sup> |
| COPM-S   | 3.646      | 3.667      | 0.973 <sup>c</sup> |

MACS=Manual Ability Classification System, AHA=Assisting Hand Assessment, Melbourne=Melbourne Assessment of Unilateral Limb Function, COPM-P/ COPM-S=Canadian Occupational Performance Measure (P=performance, S= Satisfaction).

<sup>a</sup> Chi2 test, <sup>b</sup> Mann Whitney U test, <sup>c</sup> t-test

Reference values are available for adults and children older than six years. Recently, the test-retest reliability of the Box and Block test was studied in typically developing children aged 3 to 10 years and was found to be good (ICC=0.85).<sup>22</sup> Only the affected side of the children in the mCIMT-BiT group was tested with the Box and Block test, while the nonaffected hand remained constrained. A 15-second practice trial was performed before the actual test started.

### **Long-term effects**

Primary outcome measures for the assessment of long-term effects were the AHA, evaluating the spontaneous use of the affected hand in a semi-structured observation, and the ABILHAND-Kids, evaluating manual skills through a parent's questionnaire. Secondary outcomes were the Melbourne Assessment of Unilateral Upper Limb Function

(Melbourne),<sup>23</sup> evaluating (unilaterally) the quality of upper limb movements, and the Canadian Occupational Performance Measure (COPM).<sup>24</sup> The COPM determined the parents' scores on the five most important daily life problems that their child experienced in relation to bimanual activities. For the children aged 2.5 to 5 years we used the draft version of the Melbourne that was modified specifically for this younger age group, whereas the original Melbourne was used for the older children. At the one-year follow-up assessment of the mCIMT-BiT group, only the AHA and ABILHAND-Kids were administered.

### Data and statistical analysis

To analyze the learning curves, the scores on the Box and Block test were individually examined for each participant in the mCIMT-BiT group. The instant of the maximum training effect was defined as the moment the top of the learning curve was reached. This top was independently determined by two raters through visual examination of the absolute scores. In the case of more than one top, the first top in the learning curve was taken. Subsequently, the data set was split in children that had reached a maximum effect within the six weeks mCIMT period and those who had not. These two groups were then compared with regard to demographic (age, gender) and clinical (affected side, MACS score) characteristics (using  $\chi^2$ -test for dichotomous variables and Mann-Whitney-U test for the MACS). In the case of a group difference for any of these variables, the influence of this variable on reaching the top of the motor-learning curve within the six weeks mCIMT period was expressed in a relative risk (RR) as well as in an Odds ratio (OR) with accompanying 95% confidence intervals. Group differences in outcome measures were tested with independent t-tests.

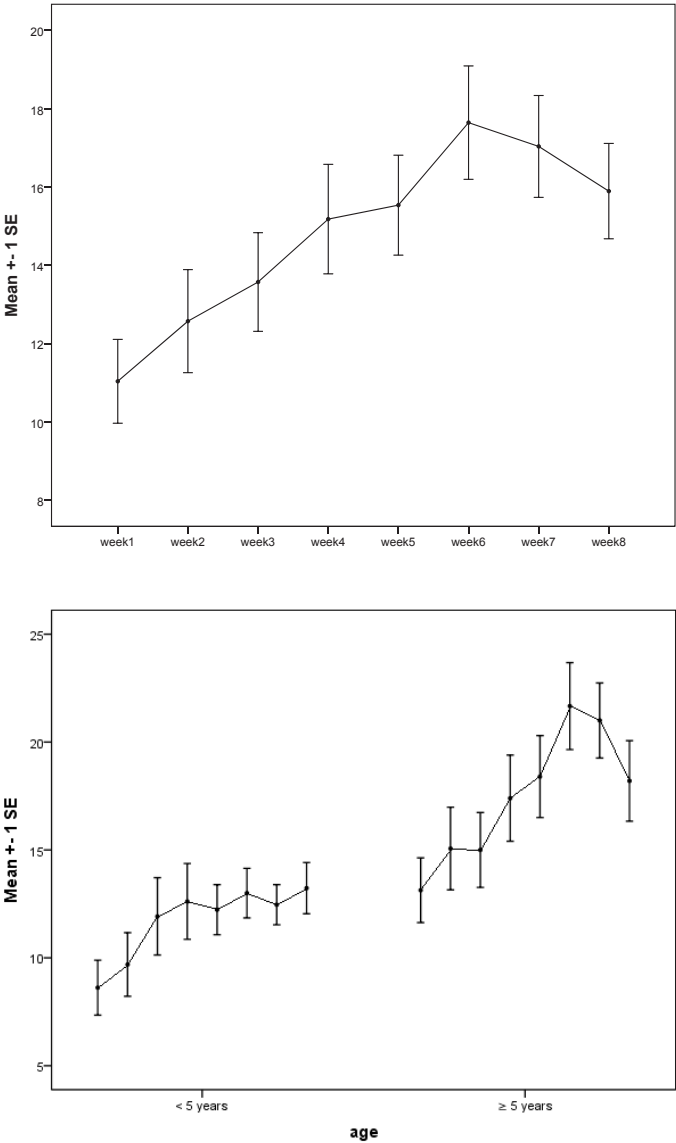
To determine the long-term effectiveness, we performed a repeated measures ANOVA of Time (one week, two months, six months, and (only for the primary outcomes in the mCIMT-BiT group) one year post intervention) for each group separately. In the case of a significant time effect, post-hoc pair-wise comparisons with Bonferroni correction were performed. All original outcome measures were handled as interval data (AHA (logits), ABILHAND-Kids (logits), Melbourne and COPM). Analyses were based on the intention-to-treat principle.

## Results

### Learning curve

No disagreement occurred between raters. In two children, more than one top was observed in the learning curve, in which cases the first top was taken as the instant of maximum training effect. The 28 children in the mCIMT-BiT group together showed a top performance on the Box and Block test at six weeks after the start of the mCIMT. Thereafter, the average unilateral performance curve declined during the two weeks of BiT (figure 1A).

**Figure 1** Weekly results box and block tests; **A.** Total sample and **B.** Sample split in 2 age groups



On the Y-axis the number of blocks; a higher number reflecting better manual dexterity.

Of these children, 15 (53.6%) had truly reached a maximum training effect between two and six weeks of mCIMT (on average at five weeks post training onset), whereas 13 children (46.4%) reached a maximum training effect during the BiT period (on average at seven weeks post training onset). These groups tended towards a significant difference with respect to age ( $p=0.064$ ). Gender, affected side and MACS score did not influence the moment of reaching a maximum training effect. Children younger than five years of age had a greater chance to reach a maximum score within the six weeks mCIMT period than children aged five years and older ( $RR=2.31$ ,  $95\%CI=1.06-5.01$ ;  $OR=6.67$ ,  $95\%CI=1.24-35.71$ ).

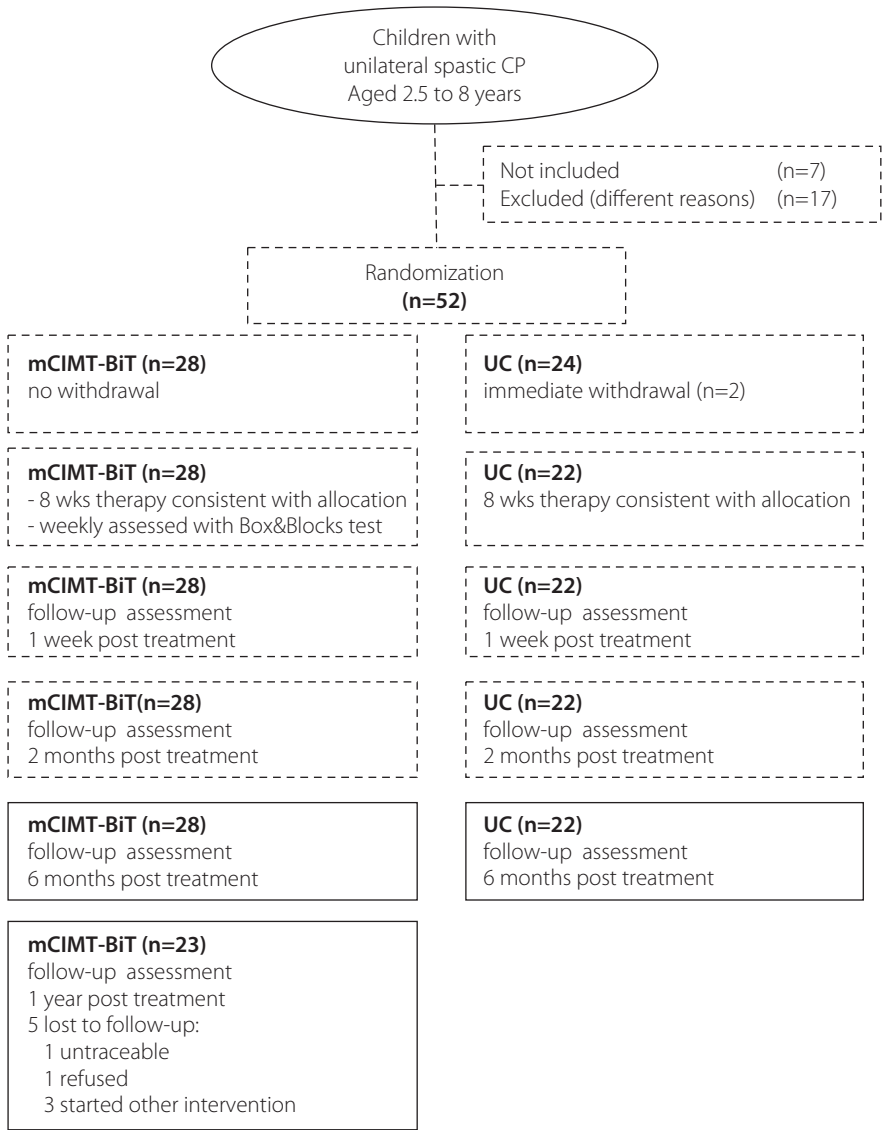
Table 2 represents the characteristics of the mCIMT-BiT group split into the two age groups. The age range of the participants was dichotomized into  $< 5$  years and  $\geq 5$  years, creating two nearly equal groups, consistent with the age distinction of the AHA versions (i.e. small kids AHA and school kids AHA). Seventy-seven percent of the younger children reached their top within six weeks and 54% even within five weeks. In contrast, 67% of the older children reached their highest score in the seventh or eighth week during the bimanual training phase. Figure 1B shows a higher mean score at onset ( $p=0.033$ ) and a noticeably longer learning curve for the older children. In addition, it becomes clear that the overall decline in Box and Block performance during the BiT period was fully attributable to the oldest age group. The amount of improvement on the Box and Block test during the mCIMT period was greater for the older children (mean difference in gain was 4.15,  $p=0.002$ ). However, at the end of the BiT period, the Box and Block test no longer showed differences in functional gain between the age groups (mean difference 0.45,  $p=0.724$ ), nor did any of the original primary or secondary outcome measures (table 2).

### Long-term effectiveness

There was no loss to follow-up in either the mCIMT-BiT or UC group at six months post intervention. At one year, five participants of the mCIMT-BiT group were lost to follow-up: one was untraceable, one refused re-assessment, and three children were not re-assessed because they had started another intervention. The patient flow is shown in figure 2.

In table 3 the raw data of all outcomes are presented, with previously published data in grey. We added the logit scores of the AHA and ABILHAND-Kids to the scaled scores and raw scores, respectively, that were presented in the original publication.<sup>1</sup> None of the outcome measures in either group showed a main effect of Time, except the Melbourne in the mCIMT-BiT group ( $F_{(1.66,44.69)}=6.456$ ,  $p=0.006$ ). Post-hoc pair-wise comparisons showed that in this group the Melbourne had significantly improved between one week and six months post intervention ( $+2.98$ ;  $95\%CI=0.16-5.80$ ,  $p=0.036$ ). Generally, long-term retention of effects did not differ between children younger than 5 years and those who were older.

**Figure 2** Patient flow RCT



Boxes with interrupted lines reflect the design of the previous published study.

## Discussion

The goal of this study was to investigate the progression of manual dexterity using the Box and Block test *during* a six-week mCIMT program followed by a two-week BiT program that has proven to be effective in children with unilateral spastic CP aged 2.5-8 years. We aimed to determine whether and when a maximum training effect was reached and what factors might influence the learning curve. In addition, follow-up data of the original outcome measures were reported to determine long-term effectiveness.

### Learning curve

The most cardinal finding of the present study was that age had a major effect on the speed of dexterity gain with the affected upper extremity during the mCIMT period. Compared to older children, children younger than five years had a 2.3 times greater chance of reaching a maximum performance on the Box and Block test within six weeks of intensive unilateral training. These children most clearly improved during the first three to four weeks of training, after which they approached a stabilization phase at five or six weeks post onset. In contrast, the children of five years and older clearly improved their Box and Block performance until the six<sup>th</sup> week of mCIMT, after which they showed a decline in performance during the BiT period. This pattern of results is indicative of the notion that the older children, on average, did not yet reach their maximum capacity level. They certainly did not reach a stabilization phase during or after the six weeks of mCIMT.

A possible explanation for the lack of a 'tapering off' during mCIMT might be that older children need more time for the cognitive phase of motor learning, whereas younger children progress faster to the associative phase.<sup>25</sup> In older children compensatory grasp and release strategies usually exist longer, as a result of which it may take more time to replace these by more appropriate strategies. Thus, older children might have benefitted from a longer mCIMT period than six weeks. This notion is supported by a review<sup>15</sup> of several small-sized studies in search of the appropriate frequency and ingredients of CIMT. This review suggested that older children are likely to need more intensive training than younger children. This notion is supported by the observation that the older children in our study showed a functional decline in unimanual dexterity during the bimanual training phase. It is unlikely that this decline can be attributed to the bimanual training itself, as studies comparing CIMT and bimanual training<sup>6,7</sup> clearly demonstrated that both interventions have positive effects on dexterity of the affected hand in children with CP (as measured with the Jebson Taylor test). Interestingly, the shorter learning curve in the younger children during the unimanual training coincided with less functional gain on the Box and Block test. However, after the BiT period, there were no longer differences between age groups in terms of Box and Block performance, spontaneous hand use (AHA), manual skills in daily activities (ABILHAND-Kids), or quality of upper limb movements (Melbourne). This finding is coherent with previous studies<sup>6,26</sup> showing that the net result



of CIMT is generally not age dependent. Still, the data of the present study suggest that, while a six-weeks training period (incorporating 54 hours of mCIMT) is sufficient for children younger than five years to reach a maximum unimanual capacity and show sufficient retention during a subsequent bimanual training phase, children of five years and older may profit from a longer period of mCIMT to reach their maximum unimanual and retain this level during subsequent bimanual training.

Next to age, we could not identify any other factors influencing the learning curve during mCIMT. In particular, we found no influence of gender, affected body side or initial level of manual ability. Children classified as MACS I transported the highest number of blocks while children classified as MACS III transported the lowest number, but their respective learning curves were almost similar. Apparently, children with poor manual ability at baseline have at least equal potential to benefit from mCIMT as children with high initial manual ability.<sup>16</sup>

### **Long-term effectiveness**

All outcome measures of the original study remained at least stable in both groups at six months, while the primary outcomes in the mCIMT-BiT group were stable even until one year after the intervention. Retention of effects at six months post intervention was not only found for the AHA and ABILHAND-Kids, but also for the satisfaction and performance scores of the five most important daily life problems as assessed with the COPM. These findings are coherent with the results of the INCITE trial,<sup>10</sup> comparing CIMT with bimanual training. Using a total dosage of 60 hours of CIMT in two weeks time, this study reported that the gains for CIMT found on the AHA, Melbourne and COPM were retained at six months and one year after the intervention. Another study, comparing CIMT and functional bimanual training (HABIT),<sup>6</sup> using a total dosage of 90 hours therapy in three weeks, reported retention of effects on the AHA and Jebson-Taylor test at sixth months follow up. Although the present study used a different protocol, it confirms the retention of results observed in the CIMT arm of the INCITE trial.<sup>10</sup> Compared to the CIMT/HABIT study,<sup>6</sup> our study demonstrates that a similar level of retention can be reached with a shorter period of CIMT than 90 hours.

As in the INCITE trial, we found that the quality of movement of the affected upper limb in the mCIMT-BiT group, as assessed with the Melbourne, showed a statistically significant and substantial further improvement at six months compared to one week post intervention, whereas the UC group showed a smaller, gradual improvement on the Melbourne only until two months after the intervention period. The question is whether the post-intervention improvement of manual skills in the mCIMT-BiT group can be attributed to the training.

Klingels et al.<sup>27</sup> studied the evolution in arm and hand function in children with unilateral CP, who were on average 9.1 years old, and found no significant spontaneous time effects on the Melbourne, AHA, or ABILHAND-Kids in a one-year period. Even though

**Table 3** Outcome measures from baseline to follow-up for the mCIMT-BiT and UC groups

|                       | mCIMT-BiT |               | UC    |                |
|-----------------------|-----------|---------------|-------|----------------|
|                       | mean      | 95% CI        | mean  | 95% CI         |
| AHA scaled score      |           |               |       |                |
| baseline              | 53.25     | 47.40 - 58.90 | 50.55 | 40.58 - 60.51  |
| 1 week                | 60.07     | 54.13 - 66.02 | 53.05 | 43.22 - 62.88  |
| 2 months              | 59.68     | 54.45 - 64.91 | 52.32 | 42.82 - 61.81  |
| 6 months              | 60.00     | 55.32 - 64.68 | 54.05 | 43.89 - 64.20  |
| 1 year                | 61.74     | 55.12 - 68.35 |       |                |
| AHA logit scores      |           |               |       |                |
| baseline              | .572      | -.265 - 1.411 | .177  | -1.248 - 1.602 |
| 1 week                | 1.640     | .834 - 2.446  | .551  | -.855 - 1.957  |
| 2 months              | 1.443     | .576 - 2.310  | .390  | -.997 - 1.779  |
| 6 months              | 1.663     | 1.015 - 2.310 | .623  | -.779 - 2.027  |
| 1 year                | 1.883     | .963 - 2.802  |       |                |
| Abilhand              |           |               |       |                |
| baseline              | 20.86     | 18.90 - 22.82 | 22.55 | 19.51 - 25.58  |
| 1 week                | 28.36     | 26.06 - 30.65 | 23.68 | 21.04 - 26.33  |
| 2 months              | 28.86     | 26.83 - 30.88 | 24.36 | 21.45 - 27.27  |
| 6 months              | 27.04     | 24.11 - 29.96 | 25.27 | 22.20 - 28.35  |
| 1 year                | 30.75     | 28.74 - 32.39 |       |                |
| Abillhand logit score |           |               |       |                |
| baseline              | .107      | -.247 - .461  | .427  | -.121 - .975   |
| 1 week                | 1.394     | .954 - 1.835  | .650  | .174 - 1.135   |
| 2 months              | 1.646     | 1.215 - 2.077 | .599  | .060 - 1.138   |
| 6 months              | 1.407     | .875 - 1.940  | .924  | .308 - 1.539   |
| 1 year                | 1.831     | 1.450 - 2.212 |       |                |
| Melbourne             |           |               |       |                |
| baseline              | 63.9      | 58.9 - 68.7   | 62.1  | 54.9 - 69.3    |
| 1 week                | 68.8      | 64.3 - 73.3   | 63.5  | 56.1 - 70.9    |
| 2 months              | 69.1      | 64.4 - 73.8   | 65.1  | 58.8 - 71.4    |
| 6 months              | 71.8*     | 67.7 - 75.9   | 65.0  | 58.4 - 71.6    |
| COPM - P              |           |               |       |                |
| baseline              | 2.9       | 2.5 - 3.4     | 3.4   | 2.9 - 3.9      |
| 1 week                | 6.5       | 6.0 - 6.9     | 4.6   | 4.0 - 5.2      |
| 2 months              | 6.5       | 6.1 - 6.9     | 4.7   | 4.1 - 5.3      |
| 6 months              | 6.5       | 6.0 - 7.0     | 5.0   | 4.5 - 5.7      |
| COPM - S              |           |               |       |                |
| baseline              | 3.7       | 3.3 - 4.1     | 3.9   | 3.4 - 4.4      |
| 1 week                | 7.4       | 6.9 - 7.8     | 5.2   | 4.7 - 5.8      |
| 2 months              | 7.3       | 6.9 - 7.8     | 5.5   | 4.9 - 6.0      |
| 6 months              | 7.3       | 6.8 - 7.8     | 5.4   | 4.6 - 6.1      |

\* significantly different from one week post treatment

Grey numbers have been published previously

AHA= Assisting Hand Assessment, Abilhand= ABILHAND-Kids, Melbourne= Melbourne Assessment of Unilateral Upper Limb Function, COPM-P/ -S=Canadian Occupational Performance Measure (P=performance, S= Satisfaction)

the children in the present study were on average younger, it is not likely that the observed delayed improvements on the Melbourne are attributable to natural development in a relatively short time period of 6 months. Indeed, Hanna et al.<sup>28</sup> have reported that children with spastic CP attain a maximum quality of upper-extremity movement at an average age of 46 months assessed with the Quality of Upper Extremity Skills Test (QUEST) and at the age of 60 months when assessed with the Peabody Developmental Motor Scale (PDMS). Thereafter, they show a tendency to performance decline depending on body-site distribution and severity of impairments. Thus, we believe that the post-intervention improvement on the Melbourne in the mCIMT-BiT group should be attributed to a delayed therapy effect rather than natural development. This delayed effect can perhaps be explained by the increment in degrees of freedom as movement coordination advances by further mastering the learned skills.<sup>29</sup> This improved motor coordination is particularly reflected in the items of the Melbourne concerning fluency and range of motion.

### **Study limitations**

The learning curve data was based on a relatively small number of participants, which may have affected the statistical power to find clinically relevant determinants of motor learning other than age. The instrument (i.e. Box and Block test) that was selected to monitor the motor learning process has recently been validated for children from the age of three.<sup>22</sup> Nevertheless, the authors of this study emphasized that for children of three years sufficient motivation and clear instructions are important to obtain reliable results. Although 10.7% of the present study sample was between 2,5 and 3 years of age, we experienced that all children were well able to complete the test in our study. Another consideration is that the Box and Block test may show a small practice effect across repeated assessments,<sup>22</sup> which has to be taken into account when interpreting the present results. Such an effect may have influenced the observed gains, but not the instants of the maximum training effects. In this study we applied the Box and Block test only to the affected hand, because the children were tested while the non-affected hand was constrained. As a result, they could not profit from the possible benefits of anticipatory action planning<sup>30</sup> by first executing the task with the non-affected hand.<sup>31</sup> Yet, this disadvantage was consistent across all assessments.

### **Conclusion**

To our knowledge, this is the first study to determine the progression of manual dexterity *during* a mCIMT intervention in children with unilateral spastic CP between 2.5 and 8 years of age. The initial level of manual ability as determined by the MACS classification did not affect the response to mCIMT, however, age was found to significantly affect the learning curve. In contrast, long-term retention of effects was not influenced by age. The majority of the children younger than five years reached a maximum and stable unimanual

performance on the Box and Block test with their affected arm and hand well within six weeks (54 hours) of training, whereas older children did not. Because not all younger children reached a maximum capacity within six weeks, diminishing the mCIMT dosage is not recommended for this group. On the other hand, the present data give support to the notion that children of five years and older might profit from more than 54 hours of mCIMT to reach their maximum unimanual capacity and retain this level during subsequent bimanual training. Future studies should specifically focus on these older children to establish the optimal dosage of mCIMT.

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# 4

Intensive upper limb intervention  
with self-management training  
is feasible and promising  
for older children and adolescents  
with unilateral cerebral palsy

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## Abstract

This study explored the feasibility and preliminary effectiveness of a short (one week) intensive intervention combining Constraint Induced Movement Therapy (CIMT) and bimanual training (BiT) to improve upper limb capacity and bimanual performance guided by individual goal setting in children and adolescents with unilateral Cerebral Palsy aged 8-18 years. Self-management training was added to the intervention to maximize the effect of training and to empower the participants in self-monitoring the effective use of their affected hand. Functional goals (Canadian Occupational Performance Measure), unimanual capacity (Box and Block test), bimanual performance (ABILHAND-Kids, Children's Hand-use Experience Questionnaire(CHEQ)) and amount of use ( Video Observation Aarts and Aarts - determine developmental disregard (VOAA-DDD-R)) were measured at baseline, one week and four months post intervention. Twenty children (mean age 9.5 years) participated. Repeated measures ANOVA was used to measure effects over time. Compared to baseline, there were significant improvements on all outcome measures. The largest effect sizes were found for the COPM-performance and COPM-satisfaction (Cohen's  $d=2.09$  and  $d=2.42$ , respectively). The effect size was large for the ABILHAND-Kids ( $d=0.86$ ), moderate for the CHEQ ( $d=0.70$ ) and Box and Block test ( $d=0.56$ ), and small for the VOAA-DDD-R ( $d=0.33$ ). All effects were retained at the four months post intervention assessment. The results of this study indicate that one-week (36 hours) intensive CIMT-BiT combined with self-management training is a feasible and promising intervention for improving the capacity of the upper limb and its use in bimanual activities in older children and adolescents with unilateral CP.

Unilateral Cerebral Palsy (CP) is the most common type of CP in children.<sup>1</sup> It can lead to a wide variety of problems in upper limb (UL) capacities and subsequent limitations in the performance of unimanual and bimanual activities of daily life. During the last decade, intensive activity-based interventions such as Constraint Induced Movement Therapy (CIMT) and Bimanual Training (BiT) have become available as UL rehabilitation approaches. In short, CIMT involves the use of restraining the unaffected UL emphasizing unimanual practice with the affected hand, whereas BiT involves UL training through bimanual tasks. Both interventions have in common that they offer high-intensity training to improve UL capacities and achieve individual goals. There is a wide variation in the intensity and duration of these UL interventions.<sup>2</sup> The effects of both types of intervention have been studied in several RCTs and were shown to be equally effective compared to usual care.<sup>2-5</sup> Yet, there are indications that CIMT is somewhat superior to BiT with regard to unimanual capacity<sup>4</sup> and BiT superior to CIMT with regard to both individual goal attainment<sup>6</sup> and bimanual coordination<sup>7</sup>. Recently, it has been suggested that a combination of CIMT followed by BiT might offer additive value to optimize the performance level of bimanual activities.<sup>3, 4, 8, 9</sup> This sequential approach was effectuated in the 'hybrid' CIMT-BiT intervention "the Pirate group"<sup>10</sup> presuming an optimal motor learning process by starting with unimanual training (CIMT) followed by intensive and goal-directed bimanual training (BiT). This hybrid approach has shown to improve UL capacity, bimanual performance and individual goals in young children (2,5 - 8 years) with unilateral CP;<sup>10</sup> but the effects of this CIMT-BiT combination have not yet been studied in older children.

School children and adolescents (8-18 years) with unilateral CP need more time to acquire the necessary motor skills of daily life than their typically developing peers.<sup>11</sup> At this age, new challenges arise when the limited capacity of the affected UL hinders the acquisition of age-appropriate bimanual skills, such as working with a ruler, precise cutting with scissors, buttoning tight trousers, using cutlery for meat, using make-up, and cooking and baking. At the same time, parents of older children with unilateral CP often express their concern about the diminishing amount of use of the affected UL as their children gradually acquire greater skills to unimanually perform many daily activities with their unaffected hand.<sup>12</sup> This discrepancy between the need to learn new age-appropriate skills and the tendency of older children to disregard the capacity of their affected UL calls for effective UL (booster) interventions to maintain and improve both the unimanual capacity and bimanual performance of age-appropriate activities.

In contrast to younger children, the school program prevails for children eight to eighteen year of age. As a consequence, high intensity interventions must be restricted to short time periods during school holidays. To accommodate the needs of this group of children with unilateral CP, we developed a day-camp intervention that combines high intensity CIMT and BiT condensed in one week (40 hours) during school holidays. Until now, day-camps with only one type of intervention (CIMT or BiT) have been described for younger and older children with unilateral CP providing 30 to 90 hours of training during

one to three weeks. Reports were based on controlled trials studying the effects of CIMT compared to BiT<sup>3,4,13</sup> and on either controlled<sup>14</sup> or uncontrolled trials<sup>15</sup> studying the effects of BiT compared to no training. All day-camps were found to be effective with regard to improving UL capacity and performance, except a 30-hour training program in 5 days.<sup>13</sup> None of these day-camps offered a combination of CIMT and BiT. Because the exact critical dose of CIMT or BiT to achieve meaningful changes in either unimanual capacity or bimanual performance is still a matter of debate,<sup>2</sup> we decided to test whether a short combined CIMT-BiT approach would be feasible and effective to improve both unilateral UL capacity and spontaneous use of the affected UL during bimanual activities in older (8-18 years) children with unilateral CP, guided by individual goal setting. Given the limited duration of the training (one week) it was deemed essential to maximize the effect of the intervention,<sup>16,17</sup> in terms of amount of UL use, by incorporating self-management training. Indeed, for older children and adolescents, it would be in the line of their maturation to learn to self-monitor the (supportive) use of their affected UL during daily activities and become less dependent on being prompted by their parents and/or teachers. Thus, this study aimed to determine the feasibility and preliminary effectiveness of a one-week CIMT-BiT group intervention combined with self-management training to improve UL capacity and bimanual performance in children with unilateral CP aged 8 to 18 years.

## Methods

### Participants and design

All children who previously participated in studies on UL training interventions in our pediatric rehabilitation center, or who had requested information about such interventions, were eligible. To be included, children had to be diagnosed with unilateral CP, aged between 8 – 18 years, and able to walk independently. In addition, they had to be able to formulate individual goals related to relevant and age-appropriate activities involving the UL, which was determined by means of a parent and child combined Canadian Occupational Performance Measure (COPM) interview. All participants attended regular education or special education for children with physical disabilities. No formal data on intellectual ability was collected, nor did we register the presence of epilepsy. A child was excluded when it showed behavioral problems that might interfere with the group process or when it was unable to comprehend simple instructions. Participants who underwent UL surgery or botulinum toxin injections or who received any form of UL training during a 3-month period before the study entry were excluded.

A pretest-posttest design was used, with assessments scheduled at baseline as well as one week and four months post intervention. All children were tested by the same assessor (YG) at every assessment. The first post-intervention assessment was scheduled one week after treatment to allow parents and children to evaluate the performance of

**Table 1** Characteristics of the participants (n=20)

| Characteristics                             |                 |                               |
|---|-----------------|-------------------------------|
| Age   | Median<br>range | 9y, 6mo<br>8y, 2mo – 17y, 5mo |
| Gender, <i>n</i>                            | Male            | 11                            |
|   | Female          | 9                             |
| Affected side, <i>n</i>                     | Right           | 10                            |
|   | Left            | 10                            |
| MACS, <i>n</i>                              | I               | 6                             |
|   | II              | 12                            |
|   | III             | 2                             |
| Zancolli, <i>n</i>                          | 0               | 8                             |
|   | 1               | 8                             |
|   | 2a              | 4                             |
| Sensibility(affected hand), <i>n</i><br>2PD | <4mm            | 5                             |
|   | 4-7mm           | 7                             |
|   | >7mm            | 1                             |
|   | absent          | 6                             |
| Stereognosis                                | ≥11/13          | 13                            |
|   | <11/13          | 1                             |
|   | absent          | 6                             |
| GMFCS, <i>n</i>                             | 1               | 17                            |
|   | 2               | 3                             |
| Education, <i>n</i>                         | Regular         | 16                            |
|   | Special         | 4                             |

MACS= Manual Ability Classification System<sup>38</sup>

GMFCS= Gross Motor Function Classification System<sup>39</sup>

Zancolli= classification for deformity of the spastic hand<sup>40</sup>

2PD= two point discrimination measured with Disk-Criminator (n=19, one participant missing)

Stereognosis; number of correct identified items out of 13

activities at home. The assessor could not be blinded for the intervention, but had no knowledge of individual compliance to the program and received no information about changes in UL capacity or performance during the intervention. Before study entry, parents signed an informed consent to permit the use of the data for publication.

### Intervention

The group intervention took place at our pediatric rehabilitation center on five consecutive days during school holidays. During this 'day-camp' the children spent the evenings and nights at home. The intervention program consisted of a combination of CIMT (three hours a day, total 15 hours) followed by BiT (five hours a day, total 25 hours). Based on previous experience, it was decided that the amount of BiT should exceed the amount of CIMT in order to provide sufficient opportunities to train bimanual activities and

implement concurrent self-management training. Short breaks and preparatory activities such as eating, drinking and gathering, transporting and stowing away training materials were integrated in the training time. Strategies for task-specific training<sup>18</sup> as well as goal-oriented (functional) training<sup>19, 20</sup> were integrated in the intervention to improve meaningful daily life activities. During the first three hours of training (CIMT), children wore their unaffected UL in a sling and worked individually or in couples while training specific unimanual skills with their affected UL using the principles of shaping and repetitive task training. The type of skills was individually determined based on the COPM interview. Thereafter, BiT started which consisted of part-task and whole-task training with both hands, embedded in games, regular daily activities (e.g. preparing lunch, changing clothes, carrying materials), and in the activities selected as personal goals during the COPM interview. Most activities were trained in small groups (2-4 children) with individual coaching. During BiT emphasis was placed on use of the affected UL, taking into account individual UL capacity and the most efficient way of performing bimanual activities.

Self-management training was incorporated in the BiT and consisted of the components self-instruction, self-monitoring, self-evaluation and rewarding.<sup>21</sup> Before the start of each activity, children had to instruct themselves what to do and how to do it (including how to use the affected UL). Then, at random moments, children were asked to 'freeze' and -with the help of a personal coach- register whether they (1) were using the affected UL, (2) were using it in an effective manner, and (3) could do it better or differently. After completing these steps, they had to decide whether they deserved a reward (a sticker or points to be the first to finish a 'game-card'). Initially, the personal coach would give the child ample feedback, but during the week coaching was gradually reduced based on the capacity of the child to independently complete the self-management steps. An important part of the intervention was the parental instruction to reduce prompting and how to stimulate their child with regard to self-evaluation and rewarding during and after the day-camp at home.

Experienced occupational therapists (OTs) and supervised OT students, together with volunteers and other professionals, worked as trainers. All trainers followed a pre-treatment 4-hour training course and received an intervention manual for use during the day-camp. During the entire intervention program there was a 1:1 trainer to child ratio.

## **Outcome measures**

### ***Primary outcome measure***

The COPM<sup>22</sup> was selected as the primary outcome measure. It assesses both the subjective performance and satisfaction with performance of relevant but problematic activities as experienced by the child and its parents. In this study we asked the children and their parents to make a list of maximally 10 bimanual problematic activities and to rate the three most important activities with regard to performance and satisfaction with performance on a scale from 1 to 10.

### ***Secondary outcome measures***

Unimanual capacity was assessed with the Box and Block Test (BBT); a valid and reliable tool with norm scores established for children aged 3-19 years old.<sup>23,24</sup> In this test of gross manual dexterity, the child had to transport as many blocks as possible in 60 seconds from one compartment to another. In addition, the 14-item Modified House Classification (MHC) scale<sup>25</sup> was used to assess unimanual functional capacity. This scale is a Rasch-based unidimensional and hierarchic set of 14 items based on the original MHC; a valid tool for children with unilateral CP with good interrater reliability (ICC=0.93). The child was asked to use several objects and to show its best capacity to use the affected UL handling the objects. The weighted sum score was converted into a logit score (range -1.838 to 1.915 logits) that expresses the unimanual capacity to grasp, hold, manipulate and release objects.

To assess bimanual performance we chose parent(or patient)-reported outcomes that reflect the independency to execute bimanual activities at home. We used the ABILHAND-Kids Questionnaire,<sup>26</sup> which is a Rasch-based parent questionnaire focusing on a child's manual ability to execute 21 daily activities that require the use of the upper limb(s). It is a valid and reliable scale in the activity domain of the International Classification of Functioning, Disability and Health (ICF), developed for children with CP aged 6-15 years. Items are scored on a three-level scale ('impossible', 'difficult', 'easy') and the total score ranges from -6.753 to +6.684 logits. In addition, the Children's Hand-use Experience Questionnaire (CHEQ)<sup>27</sup> was used to assess bimanual performance. This Rasch-based online questionnaire ([www.cheq.se](http://www.cheq.se)) identifies whether 29 selected bimanual activities are executed independently and in what way the affected hand is used to carry out the activities. Furthermore, the CHEQ assesses how children, aged 6-18 years, experience their executive skills regarding (1) the efficacy of their grasp, (2) feeling bothered by the way their hand can be used, and (3) the time it takes to perform the activities compared to peers. After data collection, the raw scores of these three subscales for perceived experience of hand use were converted into logit scores by the developers of the CHEQ. Clinimetric properties of the CHEQ are not yet available.

To assess the amount of spontaneous hand use at home from a parental perspective, parents were asked to score each of the following three questions on a Visual Analogue Scale (VAS), ranging from 0 ('not at all') to 100 ('constantly'): (1) How often did your child use the affected hand spontaneously during the past week? (2) How often did you prompt your child to use the affected hand during the past week? (3) How often did your child noticeably use the self-management routines at home during the past week? (question 3 was only asked at the post intervention assessments). In addition to the subjective ratings by parents, developmental disregard was assessed with the revised video-observation Aarts and Aarts, module determine developmental disregard (VOAA-DDD-R).<sup>28</sup> Developmental disregard is commonly defined as the discrepancy between UL capacity and actual use of the UL during daily life activities. The VOAA-DDD-R compares the duration of use of the

affected UL in two tasks; one task ('stringing beads') demands the use of both hands while the other ('decorating a muffin') merely stimulates bimanual activity. In previous research, a difference greater than 17.2% in duration of use between these tasks was indicative of developmental disregard. Although the psychometric properties of the VOAA-DDD-R were established only for children aged 2.5-8 years, we decided to use it in this study since there is no alternative test available for the assessment of developmental disregard in older children.

**Statistical analysis**

Due to the explorative nature of this feasibility study, a formal power calculation was not performed. To determine the effectiveness of the treatment we used repeated-measures ANOVA of Time (baseline, one week post intervention, four months post intervention) with post-hoc testing (Bonferroni) to compare post intervention results with baseline. In case of a significant effect of Time, we calculated effect sizes at four months post intervention using means with pooled standard deviations, expressed in Cohen's *d*-values (small= $0.2 < d < 0.5$ , moderate= $0.5 < d < 0.8$ , large= $d > 0.8$ ). Cohen's *d* was calculated as the difference between the mean scores pre intervention and four months post intervention, divided by the pooled standard deviation.

**Table 2** Distribution of the '3 most important goals' in the COPM domains Self Care, Productivity and Leisure

|  |       |       |
|--|-------|-------|
| Self-care  |       |       |
| (Un)dress  | 25,3% |       |
| Eat/drink  | 17,7% |       |
| Other self-care<br>(hygiene, toileting, bathing, etc.) | 19,0% |       |
|  |       | 63,0% |
| Productivity   |       |       |
| Prepare food   | 10,1% |       |
| Computer   | 3,8%  |       |
| School   | 2,5%  |       |
|  |       | 16,4% |
| Leisure  |       |       |
| Hobby  | 8,9%  |       |
| Sports   | 7,6%  |       |
| Other  | 5,1%  |       |
|  |       | 20,6% |
|  | Total | 100%  |

## Results

### Participants and adherence

Twenty children with unilateral CP participated in this study. Their demographic and clinical characteristics are presented in Table 1. Two participants were lost to follow up at four months post intervention. Another child could not attend the one week post-intervention assessment, because she broke her non-affected arm on the playground at school shortly after the day camp. As a result only the COPM, CHEQ and ABILHAND-Kids were registered for her at this point in time. Table 2 depicts the distribution of the three most important goals selected by the 20 children and their parents. The majority of these goals was related to self-care activities (63%), while the remainder was related to productivity (16,4%) and leisure activities (20,6%).

All 20 children completed the entire intervention program. They trained on average 36 hours during the one week intervention. The remaining four hours were spent on instructions, feedback, and breaks between activities.

### Canadian Occupational Performance Measure

All COPM values (means and 95% confidence intervals) at baseline as well as one week and four months post intervention are presented in Table 3. Both the COPM-Performance ( $F(2,38)=41.440, p<0.001$ ) and the COPM-Satisfaction ( $F(2,38)=58.239, p<0.001$ ) showed a significant effect of Time with an increase of 3.3 and 3.9 points between baseline and four months post intervention, respectively. A change score of 2 points is commonly regarded as clinically significant, which is supported by the large effect sizes based on Cohen's *d* values.

### Secondary outcome measures

All secondary outcomes are presented in Table 3. The BBT values revealed that the efficiency to grasp, hold and release significantly increased over time as the affected hand transported more blocks per minute ( $F(2,38)=17.230, p<0.001$ ). This 'moderate' effect was reached one week post intervention and was retained at four months post intervention. The 14-item MHC scale merely showed a 'small' effect of Time; positive post-intervention changes in functional capacity cumulated to a significant effect at four months post intervention ( $F(2,36)=6.079, p=0.005$ ).

Manual ability, measured with the ABILHAND-Kids, increased significantly across time ( $F(2,38)=8.862, p=0.001$ ) with a large effect size at four months post intervention compared to baseline. Before the intervention 62% (SD 14.9) of the activities of the CHEQ were already mastered independently and the participants executed a large amount (80.8 %) of these activities bimanually with the affected hand using grip. Nevertheless, there was an improvement in 'independent execution' ( $F(2,38)=12.799, p<0.001$ ) four months after the intervention (10,3%). There were also significant changes and moderate to small effect



**Table 3** Primary and secondary outcome measures for both groups

|  | Baseline<br>[95% CI]  | Mean scores<br>1 week post<br>intervention<br>[95% CI] | 4 months post<br>intervention<br>[95% CI] | ANOVA of Time<br>( <i>p</i> value) | Effect size<br>baseline-<br>4 months post<br>Cohen's <i>d</i> |
|--|-----------------------|--|---|------------------------------------|---|
| <b>Primary outcome measure</b>               |                       |  |   |                                    |   |
| COPM-Performance                             | 3.33 [2.78 - 3.88]    | 6.87 [6.32 - 7.41]                                     | 6.65 [5.75 - 7.54]                        | F(2,38)=41.44 (<0.001)             | 2.09 (large)  |
| COPM-Satisfaction                            | 3.29 [2.74 - 3.83]    | 7.44 [6.88 - 7.99]                                     | 7.22 [6.29 - 8.14]                        | F(2,38)=58.24 (<0.001)             | 2.42 (large)  |
| <b>Secondary outcome measures</b>            |                       |  |   |                                    |   |
| Unilateral capacity                          | 25.3 [20.7 - 29.9]    | 30.5 [25.5 - 35.5]                                     | 31.0 [26.0 - 36.0]                        | F(2,38)=17.23 (<0.001)             | 0.56 (moderate)   |
| 14-item MHC scale (logits)                   | 0.46 [0.20 - 0.74]    | 0.52 [0.21 - 0.82]                                     | 0.67 [0.37 - 0.97]                        | F(2,36)=6.08 (0.005)               | 0.33 (small)  |
| Performance                                  | 2.52 [1.99 - 3.06]    | 3.30 [2.67 - 3.93]                                     | 3.644 [2.97 - 4.32]                       | F(2,38)=8.86 (0.001)               | 0.86 (large)  |
| CHEQ independent<br>(% activities)           | 66.2 [59.2 - 73.2]    | 77.2 [71.6 - 82.9]                                     | 76.6 [69.7 - 83.4]                        | F(2,38)=12.80 (<0.001)             | 0.70 (moderate)   |
| Grip Efficacy (logits)                       | 58.86 [52.67 - 65.04] | 66.19 [59.21 - 73.18]                                  | 66.78 [58.35 - 75.22]                     | F(2,38)=5.57 (0.008)               | 0.50 (moderate)   |
| TimeTaken (logits)                           | 46.16 [39.66 - 52.67] | 51.89 [44.78 - 59.00]                                  | 55.84 [47.57 - 64.12]                     | F(2,38)=13.44 (<0.001)             | 0.61 (moderate)   |
| Feeling Bothered (logits)                    | 51.07 [44.98 - 57.16] | 58.00 [49.99 - 66.00]                                  | 58.33 [50.10 - 66.56]                     | F(2,38)=7.93 (0.001)               | 0.47 (small)  |
| Amount of use                                | 60.3 [49.8 - 70.8]    | 68.9 [61.3 - 76.4]                                     | 67.5 [57.4 - 77.6]                        | F(2,38)=5.107 (0.011)              | 0.33 (small)  |
| VOAA muffin duration<br>(VAS-scale)          | 47.7 [39.9 - 55.4]    | 60.9 [53.1 - 68.7]                                     | 58.0 [47.6 - 68.3]                        | F(2,38)=6.60 (0.003)               | 0.53 (moderate)   |
| Spontaneous use<br>(VAS-scale) Prompt to use | 46.9 [36.0 - 57.9]    | 52.4 [43.5 - 61.3]                                     | 46.2 [36.1 - 56.3]                        | F(2,38)=1.20 (0.313)               |   |

CI= Confidence Interval, COPM= Canadian Occupational Performance Measure, BBT= Box and Block Test, MHC= Modified House Classification, ABILHAND-Kids = ABILHAND-Kids Questionnaire, CHEQ= Children's Hand-use Experience Questionnaire, VOAA= revised Video-Observation Aarts and Aarts, VAS= Visual Analog Scale

sizes for the three CHEQ scales expressing 'grasp efficacy' ( $F(2,38)=5.57, p=0.008$ ), 'time taken' ( $F(2,38)=13.44, p<0.001$ ), and 'feeling bothered' ( $F(2,38)=7.93, p=0.001$ )

Parents VAS scores on the question about 'the amount of spontaneous use of the affected upper-limb' showed a significant increase over time ( $F(2,38)=6.601, p=0.003$ ) that was reached one week post intervention. The scores for the 'amount of prompting by the parents to use the affected limb' did not change across time ( $F(2,38)=1.199, p=0.313$ ). At one week post intervention the self-management routines shown by the child were rated with a mean of 52, indicating 'intermediate use', and maintained at this level four months post intervention.

The VOAA-DDD-R results showed that the duration of use of the affected hand during the muffin task increased significantly across time ( $F(2,38)=5.107, p=0.011$ ), whereas the duration of use during the beads task did not change. Prior to the intervention we identified eight children with developmental disregard. Four months after the intervention, there was no indication of developmental disregard in four of these eight children.

### Post-hoc analyses

Post-hoc analyses of all outcome measures (except the 14-item MHC scale) indicated that, in the case of significant time effects, significance was obtained between baseline and one week post intervention, while the scores remained stable thereafter. The 14-item MHC scale yielded no significant post-hoc tests, indicating that the significant effect of Time gradually occurred both during and after the intervention.

## Discussion

This study explored the feasibility and effectiveness of a short (one week) intensive intervention, combining Constraint-Induced Movement Therapy (CIMT) and Bimanual Training (BiT), to improve unilateral upper limb (UL) capacity and bimanual performance in older children and adolescents with unilateral cerebral palsy (CP) aged 8-18 years. The intervention was based on individual goal setting and integrated self-management training to obtain long-lasting effects, despite its limited duration, taking into account the time constraints for these children related to school obligations. The self-management training was a unique component of our training program and aimed to enhance the participants' awareness of the effective use of their affected UL during bimanual activities and to diminish their dependency on prompting by adults. The results showed that the participants not only satisfactorily attained their individual goals, but also improved the capacity of their affected UL and its use in bimanual activities one week after the intervention. Importantly, all beneficial effects were retained four months post intervention.

The primary and most important outcome was the improvement in the performance of activities selected by the children and their parents based on the COPM. Most of the individual goals were related to self-care activities (63%) and many of the participating

children stated that acquiring the ability to perform or improve the performance of these activities would yield specific gains such as “be as quick as others changing after sports” and “be able to eat at a friend’s place without needing help cutting my food”. This demonstrates their (a priori) focus on being able to participate in social activities. Thus, attaining their individual goals would enhance their ability to participate in daily life activities with peers. The high COPM change scores for performance (+3.3 points) and satisfaction (+3.9 points) four months after the intervention reflected the improvements with regard to performing daily life activities and social participation. Remarkably, these improvements were even larger than those reported previously after 30 to 90 hours CIMT or BiT day-camps.<sup>6, 13</sup> The efficacy of our 36-hour intervention might be explained by the fact that we not only combined CIMT and BiT, providing opportunities to practice newly gained capacities in functional daily activities, but also added self-management training, which made participants more aware and responsible of how they used their affected hand. Finally, our participants were directly involved in the goal-setting process, selecting the three most important goals they wanted to work on, which probably promoted their motivation to accomplish the goals.

The effect of our intervention on unimanual capacity was measured with dexterity tests and was found to be small to moderate. Improving speed and accuracy of grasping, holding and releasing objects, as assessed with the BBT and the 14-item MHC, was an important part of our training intervention. However, the exercises in many children were also focused on improving grip strength, active supination, and on increasing mobility and strength around the affected shoulder and elbow. Specifically these latter aspects were not captured by the selected dexterity test, which may add to the modest effect size observed for the capacity of the affected UL. The effects of our intervention on bimanual performance showed moderate to large effect sizes. Even though performance scores were already high at baseline (on average 2.524 logits on ABILHAND-Kids and independent execution of 66% of CHEQ activities), the children significantly improved their execution of bimanual activities up to four months post intervention. Change scores for independent bimanual execution of activities in the CHEQ were similar to those after a 60-hour BiT intervention in CP children of the same age range.<sup>15</sup> Importantly, the CHEQ revealed that our participants not only felt more positive about their performance of bimanual activities, but also that they needed less time to execute the activities, improved the efficacy of their grasp, and felt less bothered to use their affected hand during the activities. These improvements may have positively affected the retention of the observed effects. The fact that we found larger effect sizes for individual goal achievement and bimanual performance than for unilateral UL capacity may reflect the CIMT-BiT ratio (3:5) as it was applied in our intervention. Promoting functional improvement by incorporating a goal-oriented approach, especially during the BiT period, might have had an additional effect. Indeed, a previous study<sup>6</sup> found that practiced goals improved more than unpracticed goals, after CIMT as well as after BiT.

Although the amount of spontaneous use of the affected UL increased significantly when measured with the VOAA-DDD-R and children spontaneously used their affected hand to a greater extent at home based on parental report, the parents also reported that they kept prompting their child as often after the intervention as they did before. It remained unclear why the parents continued their prompting even though they observed increased spontaneous hand use in their children. In the light of the development of home-based programs the issue of prompting warrants further study.

The integration of self-management techniques during BiT was an important novel aspect of our intervention. We hypothesized that the amount of parental prompting would decrease, which is essential because prompting puts an extra burden on the parent-child interaction,<sup>29</sup> especially during puberty. Empowering adolescents by giving them the responsibility to monitor the use of the affected hand themselves is, therefore, a sensible treatment strategy. At the moments that activities were 'frozen', most participants were able to evaluate whether they were using their affected UL and whether they were using it effectively, but they needed feedback from their coach to reflect on how they could use their affected hand better or differently. Only a few older children were able to perform all self-management steps independently, so that coaching could be minimized. Hence, future studies should improve the efficacy of our self-management strategy and identify factors (e.g. age) that might influence this efficacy.

Limitations of this work are the relatively small sample size and the lack of a control group receiving a different intervention or usual care, which is inherent in an exploratory feasibility study. The selected outcome measures of this study do not match the main outcomes used in many other CIMT and BiT studies, i.e. the Assisting Hand Assessment (AHA), the Melbourne assessment of Unilateral Upper Limb function (MUUL), and the Jebsen-Taylor test (JTTHF), which limits the comparability of our results to those of other studies. Yet, as our intervention aimed to enlarge the independency of adolescents to perform daily bimanual activities, we felt that parent (or patient)-reported outcomes were most appropriate. In addition, the adolescent version of the AHA had not yet been validated and the JTTHF was not available to us at the beginning of our study, while the responsiveness of the MUUL was considered too low.<sup>10, 30</sup> Although epilepsy and level of intellectual development<sup>31</sup> may affect motor learning and the acquisition of self-management skills in children with CP, we did not control for these possible effect modifiers. Because of the exploratory nature of the present study, we did not assess specific characteristics (type, severity, laterality) of the brain lesions in individual children either. These characteristics could possibly have influenced their response to treatment. In future studies, with the availability of brain imaging techniques<sup>32, 33</sup> and knowledge of the relationship between specific brain lesions, corticomotor projection patterns, and hand function,<sup>34-37</sup> UL interventions may be better tailored to the neurological characteristics of individual patients.

## **Conclusion**

The results of this study indicate the feasibility and preliminary effectiveness of a 36-hour CIMT-BiT training program, incorporating self-management training, on various aspects of unimanual and bimanual performance in older children and adolescents with CP. Yet, they do not provide definitive information about the minimal amount of UL training to achieve clinically relevant results. Future research is warranted to investigate what CIMT-BiT ratio would be optimal and whether even less than 36 hours of CIMT-BiT might be effective, when combined with self-management training, compared to CIMT or BiT alone.

## **Conflicts of interest**

The authors have no conflict of interests to declare.

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# Part two

Upper limb assessment in  
unilateral CP



# 5

## Assessment of upper limb capacity in children with unilateral cerebral palsy: construct validity of a Rasch-reduced Modified House Classification

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## Abstract

To test and improve the unidimensionality and item hierarchy of the Modified House Classification (MHC) for the assessment of upper-limb capacity in children with Unilateral spastic Cerebral Palsy (UCP) using Rasch analysis. Construct validity of the Rasch-reduced item set was evaluated.

MHC items were scored from 369 videotaped assessments of 159 children with UCP (98 male; median age 6y 6mo). Construct validity was tested in 40 other children with UCP (21 male; median age 8y 2mo) by comparing total scores to the Manual Ability Classification System (MACS) and the ABILHAND-Kids.

Fifteen MHC items could be included in the Rasch analysis. The excluded items were either too easy or too difficult. Fourteen items fitted the unidimensional model ( $\chi^2=41.3$ ,  $df=39$ ,  $p=0.37$ ). The hierarchy of these items was different from the original MHC. There was a significant correlation with the MACS ( $r=-0.901$ ,  $p<0.001$ ) and the ABILHAND-Kids ( $r=0.558$ ,  $p<0.001$ ).

The original item hierarchy of the MHC can be improved in order to use its sum score for the assessment of upper-limb capacity in children with UCP. The Rasch-reduced 14-item MHC with weighted sum score shows good construct validity to measure functional capacity of the affected hand in children with UPC.

Children with unilateral cerebral palsy (CP), about 30% of the total CP population, typically present with a wide range of unilateral upper limb impairments. They have reduced capacity to handle objects with the affected hand and, consequently, are hindered in the performance of bimanual activities. To remediate these problems, various short and intensive training programmes for the upper limb in children with unilateral CP has been developed.<sup>1-4</sup> Valid evaluation of treatment effects requires assessment tools that have adequate clinimetric properties and focus on improvement of functional skills rather than on reduction of impairments. In the case of children with unilateral CP, it is important to measure treatment effects on 'performance' (i.e. the actual performance of an activity in daily life)<sup>5</sup> as well as to measure the 'capacity' (i.e. the execution of an activity in optimal conditions and standardized environment)<sup>5</sup> to use the affected hand in bimanual tasks. Several instruments are available for the assessment of upper limb capacity in children with CP. However, as yet, there is no validated instrument that specifically evaluates the maximal capacity of the affected hand to participate in bimanual tasks.

In 1981, House et al.<sup>6</sup> devised a nine-level functional classification system to describe the role of the assessed hand in children with CP as a passive or active assist in bimanual activities (Fig. 1). This classification provides categories for upper limb capacity. In 2008, Koman et al.<sup>7</sup> introduced the Modified House Functional classification (MHC) with the intention of improving the score discrimination of the original classification, to make it better suited for monitoring patients and to evaluate treatment efficacy.<sup>7</sup> Thirty-two items were added to the categories of the House classification (Fig. 1). Items were selected by means of a consensus process among experts. A sum score (i.e. number of passed items) represented the functional capacity of the assessed hand (range 0–32 points). In order to use the sum score of a scale, however, it is necessary that the scale is unidimensional (i.e. all items need to measure the same construct). Although the MHC was shown to be reproducible,<sup>7</sup> its unidimensionality or hierarchical properties have never been tested.

The Rasch model for scale validation provides a means to evaluate both the unidimensionality and item hierarchy of a scale.<sup>8</sup> One of the underlying assumptions for Rasch modelling is that items can vary from easy to difficult and, as such, can be ranked on one line, i.e. the logit unit scale (log-odds transformed probabilities). Second, individuals can be ranked from less to more able on the same scale. Thus, easy items can be performed by individuals of almost all ability levels, whereas more able persons are more likely to successfully execute the difficult items as well. Hence, in Rasch modelling, the ranked items and the item scores of the ranked subjects are compared and analyzed. Items that do not add to the unidimensional construct or hierarchy can be identified and removed from the scale. The unidimensionality and item hierarchy of the remaining item set can then be tested using model fit statistics, thus asserting that the variation in sum scores can be attributed to differences in the evaluated construct only.

**Figure 1** House functional classification system with MHC items

| House class | Designation by House      | Activity level according to House   | Hierarchically ordered MHC items |     |     |     |     |
|-------------|---------------------------|---|----------------------------------|-----|-----|-----|-----|
| 0           | Does not use              | Does not use  |                                  |     |     |     |     |
| 1           | Poor passive assist       | Uses as stabilizing weight only   | 1                                |     | 2   |     |     |
| 2           | Fair passive assist       | Can hold onto object placed in hand   | 3                                |     | 4   |     |     |
| 3           | Good passive assist       | Can hold onto object and stabilize it for use by the other hand                     | 5                                | 6   | 7   | 8   |     |
| 4           | Poor active assist        | Can actively grasp object and hold it weakly  | 9*                               | 10  | 11  | 12  | 13* |
| 5           | Fair active assist        | Can actively grasp object and stabilize it well                                     | 14*                              | 15* |     | 16* | 17* |
| 6           | Good active assist        | Can actively grasp object and then manipulate it against other hand                 | 18*                              | 19* | 20* | 21* | 22* |
| 7           | Spontaneous use, partial  | Can perform bimanual activities easily and occasionally uses the hand spontaneously | 23*                              | 24* | 25  | 26* | 27  |
| 8           | Spontaneous use, complete | Uses hand completely independently without reference to the other hand              | 28                               | 29  | 30  | 31  | 32  |

MHC= Modified House classification

\* the 14 MHC-items that fit the unidimensional model after Rasch analysis

Furthermore, the recently developed Rasch models (e.g. the One Parameter Logistic Model [OPLM] we applied) can be used to weight the items according to their discriminative capacity, asserting a balanced and valid contribution to the sum score.

In this study we sought to evaluate whether the MHC can be used as an instrument to discriminate between levels of functional capacity of the affected hand objectively in children with unilateral CP. Because the MHC was not specifically developed for children with unilateral CP, we evaluated its unidimensionality and item hierarchy in a large cohort of children with unilateral CP (the calibration cohort). Rasch analysis was used to improve both clinimetric properties by eliminating, reordering, and weighting items. The Rasch-reduced item set was then tested for its construct validity within the calibration cohort as well as in another sample of 40 children with unilateral CP (the validation cohort). We compared the weighted sum score to the Manual Ability Classification System (MACS)<sup>9</sup> and to the ABILHAND-Kids questionnaire (ABILHAND-Kids).<sup>10</sup>

## Method

### Participants

For the calibration cohort we used the videotaped assessments of all children with unilateral CP who had visited our pediatric rehabilitation department in the period September 2006 - April 2010. Children were included if (1) the assessment was videotaped and (2) it was performed and completed according to a standardized assessment protocol, and (3) written parental consent was available for use of the videotaped assessments in clinical research. This resulted in 373 videotaped assessments obtained from 159 children. All characteristics, shown in Table 1, were obtained from the assessment reports and the medical records.

A second group of 40 children with unilateral CP was included to constitute the validation cohort. These children had attended our department for modified constraint-induced movement therapy combined with bimanual training between April 2011 and February 2013 (Table 1). In this group MHC, MACS, and ABILHAND-Kids scores were assessed by an experienced occupational therapist as part of the standardized pre- and post-intervention assessments.

### Assessment of the videotapes

Scoring criteria for the various MHC items have not been published by the developers. Hence, we consulted the authors of the study by Koman et al.<sup>7</sup> to obtain the criteria they used for scoring each item as pass or fail. Six raters (five occupational therapy students and one registered occupational therapist) were instructed and trained to score the MHC items of all videotapes according to these criteria. The videotapes were blinded, so that no demographic or clinical information was disclosed to the raters.

Interrater reliability was established before the start of the study based on 10 videotaped assessments. The intraclass correlation (model 3, single measure) for the MHC sum score was 0.93 (95% confidence interval [CI] 0.84–0.98). A previous study<sup>11</sup> investigating the use of standardized videotaped examinations to establish the House classification of 10 patients with unilateral CP found interrater reliability of 0.3 and intrarater reliability of 0.58. The good agreement of our raters supports the use of recorded assessments to score the MHC in the current study.

### Rasch analysis

Rasch analysis was used to test both the unidimensionality and item hierarchy of the MHC. More specifically, we used the OPLM,<sup>12</sup> a hybrid between the one-parameter (Rasch) model and two-parameter item response theory (Birnbaum) model. First, we excluded items that were passed by less than 5% or more than 95% of the calibration cohort. The difficulty of these items could not be reliably estimated because there was too little variance in the scores. Then, we examined the unidimensionality of the sum score and



**Table 1** Demographic characteristics of the participants

|  |       | calibration cohort (n=159) | validation cohort (n=40) |
|--|-------|----------------------------|--------------------------|
| Age (median)                                   |       | 6 y, 6 mo                  | 8 y, 2 mo                |
|  | range | 2 y, 1 mo - 17 y, 5 mo     | 3y, 2 mo – 17 y, 6 mo    |
| Male   |       | 98 (62%)                   | 21 (52.5%)               |
| Affected side                                  |       |                            |                          |
|  | right | 71 (45%)                   | 20 (50%)                 |
|  | left  | 88 (55%)                   | 20 (50%)                 |
| MACS   |       | N (%)                      |                          |
|  | I     | 47 (29.6%)                 | 7 (17.5%)                |
|  | II    | 77 (48.4%)                 | 22 (55%)                 |
|  | III   | 32 (20.1%)                 | 11 (27.5%)               |
|  | IV    | 3 (1.9%)                   | 0                        |
|  | V     | 0                          | 0                        |
| Mean MHC sum score (SD)                        |       | 18.5 (5.5)                 |                          |
|  | range | 5-29                       |                          |
| House category score<br>(guided by MHC scores) |       | N (%)                      |                          |
|  | 0     | 4 (2.5%)                   |                          |
|  | 1     | 3 (1.9%)                   |                          |
|  | 2     | 11 (6.9%)                  |                          |
|  | 3     | 21 (13.2%)                 |                          |
|  | 4     | 43 (27%)                   |                          |
|  | 5     | 62 (39%)                   |                          |
|  | 6     | 9 (5.7%)                   |                          |
|  | 7     | 6 (3.8%)                   |                          |
|  | 8     | 0 (0%)                     |                          |

MACS = Manual Ability Classification System, MHC = Modified House Classification

assigned item weights, according to an item's discriminative ability, guided by the OPLM analysis. Item weights could range in discriminative ability from 1 (poor) to 5 (high) and were used to weight items before summation. Specific  $\chi^2$ -based goodness-of-fit statistics, implemented in the software, were used to examine the fit of the data to the measurement model. Data fitness tests are based on the OPLM's expected versus observed proportions of patients with a positive score on the MHC items. Misfitting items, indicated by significant item-orientated tests ( $p < 0.05$ ), were deleted, starting with the item with the highest misfit ( $\chi^2$ -value) and continuing until the overall model fit indicated unidimensionality of the MHC item set. The overall model fit was tested with the  $\chi^2$ -based R1c statistic, which  $p$ -value should exceed 0.05 to indicate that the data fit the unidimensional OPLM. MHC

item difficulties and person ability measures were determined using conditional maximum likelihood estimation and expressed on a logit unit scale, ranging from  $-3$  (easy) to  $+3$  (hard) in most practical applications. The scale is a log transformation of the odds of an MHC item being successfully carried out. Ultimately, we calculated Cronbach's  $\alpha$  of both the original MHC and the Rasch-reduced scale and compared the item hierarchy obtained by OPLM analysis with that of the current MHC.

### Construct validity

The weighted sum scores of the Rasch-reduced item set were first evaluated within the calibration cohort by relating them to the individual MACS scores and to age using Somer's  $d$  and Spearman's rho. It was hypothesized that the weighted sum scores would show a strong correlation with the MACS, a measure that classifies the ability to independently use the hands in daily activities. A low or non-existing relation with age was expected and considered important because this would confirm that the weighed sum scores were not biased by the age of a child. The correlations were then calculated for the validation cohort; the weighed sum scores with the MACS (Somer's  $d$ ) and with the ABILHAND-Kids (Spearman's rho). We expected a substantial but somewhat lower correlation with the ABILHAND-Kids than with the MACS, because the ABILHAND-Kids score for manual ability is based not only on the capacities of the affected hand, but also on the use of compensatory strategies and environmental influences.<sup>13</sup>

5

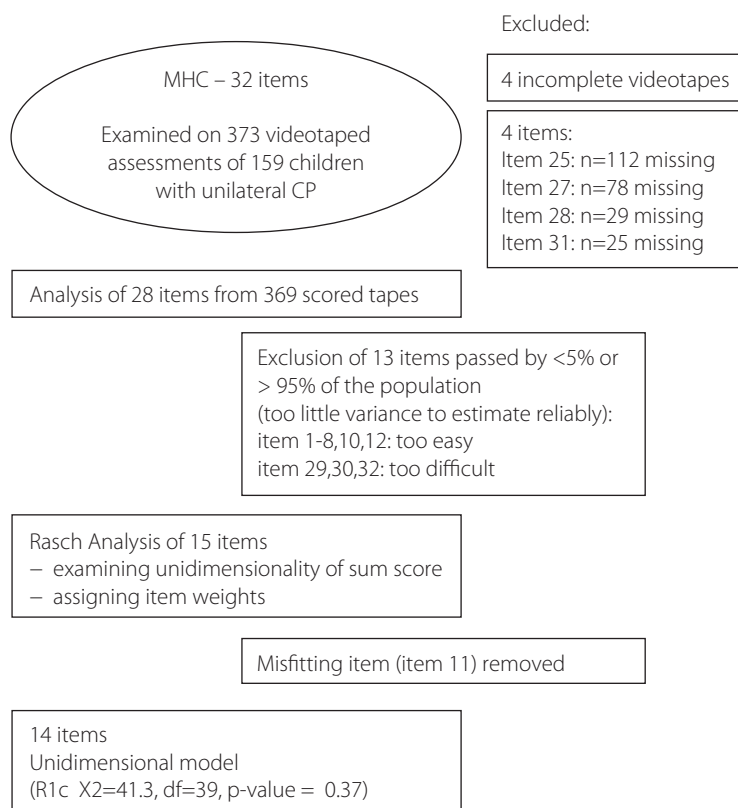
## Results

The calibration cohort comprised 159 participants (98 males, 61 females; median age 6y 6mo, range 2y 1mo–17y 5mo; Table 1). The MHC scale exhibited no floor or ceiling effects; there were no children with a maximum score and only 2.5% with a minimum score. Most children were classified at MACS levels 1 (29.6%) or 2 (48.4%) and, according to their MHC scores, were classed at House category 3, 4, or 5. A flow chart of the analysis is shown in Figure 2. In a number of cases the items 'pick up beans while holding one in ulnar side hand' (item 25), 'buttoning' (item 27), 'pick up, stabilize, and translate coins' (item 28), and 'unilaterally putting small pegs in pegboard' (item 31) were not presented in the videos. As it is most likely that these items were not assessed because they were too difficult or frustrating for the child under evaluation, we excluded them from the Rasch analysis. Indeed, when available for scoring, these items were passed in 2.2%, 27.6%, 0%, and 3.8% of the cases (items 25, 27, 28, and 31, respectively). Another 13 items were excluded from further analysis because less than 5% or more than 95% of the children passed these items. In 4 of the 373 video-assessments, values for various MHC items were missing and thus these assessments were excluded, leaving 369 video assessments and 15 MHC items for the Rasch analysis.

## Unidimensionality and hierarchy of the MHC

Table 2 summarizes the results of the Rasch analyses. The item fitness test identified one misfitting item (item 11: 'gross grasp of small block'), which was subsequently removed. The remaining 14-item set showed goodness of fit to the unidimensional model ( $R_{1c} \chi^2=41.3$ ,  $df=39$ ,  $p$ -value=0.37), indicating that the 14 items worked well together to measure a single upper limb capacity construct. Item difficulties ranged from -1.193 to 1.309 logits. The item pairs 16 and 18, 20 and 21, and 24 and 26 had similar item difficulties. The item difficulty hierarchy was different from the ordering suggested by Koman et al.<sup>7</sup> This applies specifically to items 14, 19, and 23. Based on the Rasch analysis, items 14 and 19 both had a higher item difficulty than four originally higher ranked items, whereas item 23 was

**Figure 2** Flowchart for analysis of MHC items



MHC= Modified House classification.

**Table 2** Item statistics of 14-item MHC scale. Items are ordered from easy to difficult.

| Original<br>MHC<br>Item order | Original<br>MHC<br>class | Item<br>label   | Item<br>Weight <sup>1</sup> | Item<br>Difficulty<br>( $\beta$ ) | SE<br>( $\beta$ ) |
|-------------------------------|--------------------------|---|-----------------------------|-----------------------------------|-------------------|
| 13                            | 4                        | Use body to stabilize object in hand against resistance while other hand manipulates. | 3                           | -1.193                            | .095              |
| 9                             | 4                        | Functional arm movement towards object in front.                                      | 2                           | -1.003                            | .089              |
| 17                            | 5                        | Voluntary release in space.   | 3                           | -.938                             | .077              |
| 15                            | 5                        | Rake and grasp small beans.   | 4                           | -.779                             | .070              |
| 16                            | 5                        | Retains object against moderate resistance while other hand manipulates.              | 3                           | -.499                             | .060              |
| 18                            | 6                        | Grasp/hold light resistive media without crushing while other hand manipulates.       | 2                           | -.456                             | .069              |
| 14                            | 5                        | Reach with some supination for vertically oriented object                             | 4                           | -.119                             | .046              |
| 23                            | 7                        | Bring item in hand to mouth with forearm supination                                   | 3                           | .258                              | .048              |
| 20                            | 6                        | Pad-to-pad pinch to pick up Cheerios one at the time, may have adducted thumb         | 5                           | .368                              | .038              |
| 21                            | 6                        | Point with isolated finger, other fingers out of the way                              | 4                           | .397                              | .045              |
| 22                            | 6                        | Release in space with some wrist extension  | 2                           | .450                              | .066              |
| 19                            | 6                        | Hold and turn paper in affected hand while cutting out circle                         | 3                           | .897                              | .057              |
| 24                            | 7                        | Simple rotation of object, turn ½ revolution with thumb and fingers                   | 3                           | 1.309                             | .076              |
| 26                            | 7                        | Thumb somewhat opposed in tip-to-tip pinch to pick up small items                     | 3                           | 1.309                             | .076              |

MHC= Modified House classification, SE= standard error

<sup>1</sup> To calculate a total score for the 14-item MHC scale, multiply a positive score for each item with its item weight and summate. For example, the total score for a child with a positive score on item 13, 9 and 17 equals: (1x3) + (1x2) + (1x3) = 8 points (= -.88 logit, see Table 3)

**Table 3** Conversion table of 14-item weighted sum score to obtain the functional capacity measure in logit units

| Weighted sum score | Functional capacity measure ( $\theta$ ) | Weighted sum score (continued) | Functional capacity measure ( $\theta$ ) (continued) |
|--------------------|--|--------------------------------|--|
| 0                  | -1.838                                   | 23                             | 0.139  |
| 1                  | -1.596                                   | 24                             | 0.191  |
| 2                  | -1.435                                   | 25                             | 0.240  |
| 3                  | -1.309                                   | 26                             | 0.288  |
| 4                  | -1.203                                   | 27                             | 0.334  |
| 5                  | -1.111                                   | 28                             | 0.380  |
| 6                  | -1.029                                   | 29                             | 0.427  |
| 7                  | -0.952                                   | 30                             | 0.475  |
| 8                  | -0.880                                   | 31                             | 0.525  |
| 9                  | -0.810                                   | 32                             | 0.579  |
| 10                 | -0.742                                   | 33                             | 0.639  |
| 11                 | -0.673                                   | 34                             | 0.705  |
| 12                 | -0.603                                   | 35                             | 0.781  |
| 13                 | -0.531                                   | 36                             | 0.869  |
| 14                 | -0.458                                   | 37                             | 0.967  |
| 15                 | -0.383                                   | 38                             | 1.072  |
| 16                 | -0.310                                   | 39                             | 1.182  |
| 17                 | -0.238                                   | 40                             | 1.294  |
| 18                 | -0.168                                   | 41                             | 1.413  |
| 19                 | -0.101                                   | 42                             | 1.544  |
| 20                 | -0.036                                   | 43                             | 1.701  |
| 21                 | 0.026                                    | 44                             | 1.915  |
| 22                 | 0.084                                    |                                |  |

$\Theta$ = person ability

much easier than weighted in the original MHC item ordering. The discriminative capacity of the items, as indicated by the item weights, ranged from 2 to 5. Items 9, 18, and 22 had the lowest discrimination and item 20 ('perform pad-to pad pinch') had the highest discrimination. Internal consistency (Cronbach's  $\alpha$ ) of the Rasch-reduced 14-item scale was 0.85, compared with 0.87 for the total MHC with 28 evaluated items.

### Construct validity

Within the calibration cohort the 14-item weighted sum score correlated well with the MACS ( $r=-0.688$ ,  $p<0.001$ ). There was no correlation with age ( $r=0.096$ ,  $p=0.231$ ). Thirty children (8.1%) failed the easiest item of the reduced scale and 39 (10.6%) passed the two most difficult items. Only 2.4% failed all items, whereas 2.2% passed all items and subsequently reached the maximum score. In the validation cohort the weighted sum scores of the 14-item MHC scale were approximately normally distributed with a mean of 24.65 (SD 9.8). One child passed all items (maximum score). There was a strong correlation with the MACS ( $r=-0.901$ ,  $p<0.001$ ) and a fair correlation with the ABILHAND-Kids ( $r=0.558$ ,  $p<0.001$ ). Assessment and scoring of the 14-item scale in this cohort took about 10 minutes per child.

### Discussion

The purpose of this study was to evaluate whether the MHC could be validly used to measure functional capacity of the affected hand in children with unilateral CP. We examined the unidimensionality and item hierarchy of the MHC for this particular group and tested its construct validity.

The empirical evidence demonstrated that almost half of the MHC items were either too easy or too difficult for children with unilateral CP. The Rasch analyses showed that 14 of the original MHC items fitted a unidimensional model, implying that the variation in scores can be attributed to a single construct: the functional capacity of the affected hand. Reducing the scale to 14 items did not affect the internal consistency. We found that, for some items, the item hierarchy was different from in the original scale. Furthermore, the items did not discriminate with equal precision. Hence, if they are used in a scale they need to be weighted accordingly before summation to obtain a more reliable result. Taken together, for use in children with unilateral CP, the MHC could be reordered and reduced to a 14-item scale with a weighted sum score that expresses the functional capacity of the affected hand.

Instruments that are valid to assess upper limb capacity in children with CP are scarce and the frequently used 'capacity' instruments have several limitations. The Melbourne Assessment of Unilateral Upper Limb Function<sup>14</sup> and the Quality of Upper Extremity Skills Test<sup>15,16</sup> measure (overall) upper limb quality of movement,<sup>17</sup> but have only a few items addressing grasping and releasing objects. In addition, the Quality of Upper Extremity Skills Test is not applicable to children of 8 years and older. The Jebson Taylor Hand Function Test<sup>18</sup> assesses unilateral efficiency in seven (timed) tasks focusing on unimanual capacity; however, it is a generic norm-referenced instrument and is often modified to

meet the abilities of children with CP.<sup>19,20</sup> The 14-item MHC scale specifically assesses grasping, holding, manipulating, and releasing objects. As such, this scale can fill a gap in the upper limb assessments in children with unilateral CP, because it specifically focuses on the capacities of the affected hand as an assist in handling objects.

In the calibration cohort the sum score showed a strong correlation with the MACS, confirming that children with a good MACS classification (e.g. level 1) are more likely to score high on the 14-item MHC scale. This strong convergent validity was confirmed in the validation cohort. We found that the relationship of the MHC scale with the ABILHAND-Kids was somewhat weaker, which can be explained by the characteristics of both instruments. The 14-item MHC scale assesses unilateral functional capacity, whereas manual ability assessed with the ABILHAND-Kids is a fusion of many aspects (including the capacity of the dominant hand and the use of compensatory strategies), of which the capacity of the affected hand is just one.<sup>13</sup> There was no correlation with age, suggesting that the 14-item MHC scale can validly be applied in various age groups.

The item difficulties were evenly spread over the range of the scale. With 8.1% of the children failing the easiest item and 10.6% passing the most difficult item, this 14-item MHC scale has a floor and ceiling effect, although this is not reflected in the sum score. Less than 2.5% of the calibration cohort reached a minimum or maximum score. Our study also revealed three item pairs with almost identical item difficulties. Technically it would be better to remove the less discriminative item of each pair and recalculate the effects on the properties of the scale.

## **Limitations**

The results of this study apply only to children with unilateral CP. Although our calibration cohort was typical of children with unilateral CP who are treated in rehabilitation centers, it did not represent the entire spectrum of children with CP for whom the MHC was originally developed. In particular, children with good upper limb capacity who are rarely treated in rehabilitation centers are not represented, which might account for the finding that only a few children in our study passed the items at the top of the MHC scale (i.e. items 25, 27–32), reflecting (near) normal hand function. As a consequence of the aim of study, children with bilateral CP who are more likely to have severe upper limb disabilities (e.g. MACS levels 4 and 5) were not included either, although the MHC was developed for these children as well. Future studies should, therefore, include other CP subtypes to identify the item difficulty and hierarchy of the lowest and highest MHC items.

Because this study was retrospective in nature, some data related to four items were missing from the calibration cohort and, therefore, could not be included in the Rasch analysis. We expect that the missing values were caused by these items being too difficult,

as they concerned the highest ranked items of the MHC. Although not absolutely certain, this notion was supported by the percentage of passed items in the rest of the sample; for three of the four items being scored, the pass rate was less than 4%. Item 27 ('able to fasten and unfasten a button bimanually'), for which the success rate among those in whom this item was presented was 27.6%, should be re-evaluated in future studies to establish its difficulty and contribution to the scale.

Finally, age, sex, and affected side may influence fine motor control of the upper limb in young children. Differential item functioning of the 14-item MHC scale should, therefore, be formally evaluated in various subgroups consisting of at least 200 patients each.<sup>8</sup>

## Conclusion

The results of this study suggest that the 14-item MHC scale might be useful to measure functional capacity of the affected hand in children with UCP. The reduced scale with re-ordered items has good internal consistency, but the item scores must be weighted in order to validly interpret the sum score.

Future studies are needed to further improve the applicability of the MHC scale in children with UCP. In particular, its responsiveness should be established, DIF should be tested in various subgroups, and construct validity should be strengthened by comparing the 14-item MHC scale with other well-established upper-limb measures such as the AHA and the Melbourne.

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# 6

Assessment of upper-limb capacity,  
performance, and developmental  
disregard in children with  
cerebral palsy: validity and reliability  
of the revised Video-Observation  
Aarts and Aarts module:  
Determine Developmental Disregard  
(VOAA-DDD-R)

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## Abstract

To investigate the validity and reliability of the revised Video-Observation Aarts and Aarts module Determine Developmental Disregard (VOAA-DDD-R).

Upper-limb capacity and performance were assessed in children with unilateral spastic cerebral palsy (CP) by measuring overall duration of affected upper-limb use and the frequency of specific behaviors during a task in which bimanual activity was demanded ('stringing beads') and stimulated ('decorating a muffin'). Developmental disregard was defined as the difference in duration of affected upper-limb use between both tasks. Raters were two occupational and one physical therapist who received 3 hours of training. Construct validity was determined by comparing children with CP to typically developing children. Intrarater, interrater, and test-retest reliability were determined using the intraclass correlation coefficient. Standard error of measurement and smallest detectable difference were also calculated.

Twenty-five children with CP (10 males; mean age 4y 8mo [SD 1y 7mo], range 2y 8mo to 8y; Manual Ability Classification System scores I–III) scored lower on capacity ( $p=0.052$ ) and performance ( $p<0.001$ ), and higher on developmental disregard ( $p<0.001$ ) than 46 age- and sex-matched typically developing children (23 males; mean age 5y 4mo [SD 1y 5mo], range 2y 6mo to 8y). The intraclass correlation coefficients (0.79–1.00) indicated good reliability. Absolute agreement was high, standard errors of measurement ranged from 4.5 to 6.8%, and smallest detectable differences ranged from 12.5 to 19.0%.

The VOAA-DDD-R can be reliably and validly used by occupational and physical therapists to assess upper-limb capacity, performance, and developmental disregard in children (2y 6mo to 8y) with CP.

Children with unilateral spastic cerebral palsy (CP) have motor impairments such as muscle weakness and spasticity on predominantly one side of the body.<sup>1,2</sup> These motor impairments are important causes of activity limitations.<sup>3,4</sup> According to the International Classification of Functioning, Disability and Health, the 'activity' level can be subdivided in the qualifiers 'capacity' (i.e. the execution of an activity in a standardized environment) and 'performance' (i.e. the actual performance of an activity in daily life).<sup>5</sup> Children with CP not only experience limitations in their capacity, but they also tend to underuse their affected upper limb in daily life (i.e. limited performance) given their individual capacity. This lack of spontaneous use of the affected limb in developing children is also referred to as 'developmental disregard'.<sup>6</sup>

To design an individually tailored rehabilitation program, detailed assessment of upper-limb disability is essential.<sup>7</sup> Therefore, it is important to assess bimanual activities because many children who have developmental disregard prefer to use their less-affected upper limb in unimanual tasks. They will only use their affected limb during bimanual tasks. However, tests of upper-limb use during bimanual activities are scarce,<sup>8,9</sup> and many functional measures focus on unilateral tasks.<sup>10,11</sup> Only the Assisting Hand Assessment<sup>12</sup> consists of semi-structured bimanual tasks to assess the effectiveness of use of the assisting upper limb. Although the Assisting Hand Assessment provides a summed frequency score of the effectiveness of upper-limb use, it does not assess the duration of spontaneous use. Because the overall duration of upper-limb use takes into account all motor behaviors, including (unsuccessful) attempts to involve the affected arm and hand, it seems to be a more valid indicator of developmental disregard than merely counting the frequency of successful behaviors.

To assess both the overall duration and frequency of affected upper-limb use, the Video Observations Aarts and Aarts module to Determine Developmental Disregard (VOAA-DDD) was developed.<sup>13</sup> It consists of two standardized tasks, 'stringing beads' and 'decorating a muffin', to assess upper-limb use. The beads task was designed to demand the use of both hands to accomplish the task, whereas the muffin task was designed merely to stimulate bimanual activity (the task is most efficiently performed with both hands). By using structured video observations and a custom-designed software program,<sup>14</sup> the tasks can be scored offline for the occurrence of specific motor behaviors (i.e. frequency) and the total duration of affected upper-limb use. When used by trained occupational and physical therapists, the VOAA-DDD was shown to be reliable and valid in children between 2 years 6 months and 8 years old with unilateral spastic CP.<sup>13</sup> However, the scoring system of the VOAA-DDD was very elaborate and the numbers of subtasks and repetitions were not consistent in the muffin and beads tasks.

Recently, the VOAA-DDD was revised (VOAA-DDD-R) to improve feasibility and interpretation. First, the distinction between the beads task and the muffin task was made more pronounced in terms of demanding versus stimulating bimanual hand use. Second, the beads and muffin task now have the same number of subtasks, which is also the same

for all ages. Third, the motor behaviors that need to be scored were reduced from 10 to the three most important behaviors (i.e. grasp, hold, release). These behaviors were shown to be essential to perform each subtask and did not differ in frequency between the dominant and non-dominant hand in typically developing children.<sup>15</sup> Finally, only three scores are used to reflect different aspects of upper-limb use: a capacity score (i.e. the frequency during the beads task), a performance score (i.e. the frequency during the muffin task), and a duration score (i.e. the difference in the duration of upper-limb use between the beads and the muffin task).<sup>15</sup> The last score was used as an operationalization of developmental disregard. These revisions required a new investigation of the psychometric properties of the VOAA-DDD-R. The goal of the present study was to investigate the construct validity and the intrarater, interrater, and test–retest reliability of the VOAA-DDD-R in children with unilateral spastic CP.

## Method

### Participants

Twenty-five children with CP were recruited from two rehabilitation centers in the Netherlands (Sint Maartenskliniek, Nijmegen, and Rijndam Rehabilitation Center, Rotterdam). This sample size was based on the results of our previous study. Inclusion criteria were (1) CP with a unilateral spastic movement impairment, (2) age between 2 years 6 months and 8 years, and (3) Manual Ability Classification System<sup>16</sup> scores I, II, or III. Children were excluded when they could not understand or execute simple tasks because of intellectual disability (i.e. developmental age below 2y). In addition, we recruited 46 age- and sex-matched typically developing children from two regular primary schools in Elst and Almere, the Netherlands, and one pre-school playgroup in Nijmegen, the Netherlands. Hand dominance of the comparison participants was determined based on parental information and on the hand the children used when they were asked to write their name or draw a picture. Legal caregivers provided written informed consent for all participants. All procedures in this study were approved by the regional medical ethics committee.

### Raters

Two occupational therapists and one physical therapist experienced in the treatment of children with movement disorders performed the offline scoring of the videos, for which they received training for 3 hours.

### Tasks

Both the beads and the muffin task consist of four subtasks. In the beads task (Fig. 1a), the child was asked to string beads (flat discs) on a shoelace as if to feed a caterpillar. First, the child was asked to open a closed can and to grasp a disc from the can, to place the disc on

the table, and to put the lid back on the can. Second, the child had to pick up an egg-timer, to turn it so that the timer went off (as if to wake the caterpillar), and to place the timer back on the table. Then the child had to open a drawer that was being held back by elastic bands, and take out the shoelace. Third, the child had to pick up the disc and to string it on the shoelace. Fourth, the child had to open the drawer, put back the shoelace, and pick up the egg-timer to reset it. In the muffin task (Fig. 1b), the child was asked to decorate a muffin with sweets. First, the child was asked to open a can, grasp a sweet from the can, place the sweet on a plate, and put the lid back on the can. Second, the child had to open a play oven and take out a muffin that was placed in a sieve with a handle. Third, the child had to grasp a stick that was placed upside-down in an open can and make a hole in the muffin using the stick. The child was then asked to take the sweet and to put it in the hole in the muffin. Fourth, the child had to place the sieve holding the muffin back in the oven and close the door. All subtasks were repeated four times.

The beads and the muffin task both lasted 2 to maximally 7 minutes. Participants were seated in a chair with their back supported, their forearms and hands laying on the table, and their feet placed on the floor or a footplate. In the case of a child with CP, the

**Table 1** Characteristics of the children with cerebral palsy (CP) and the typically developing children (TDC)

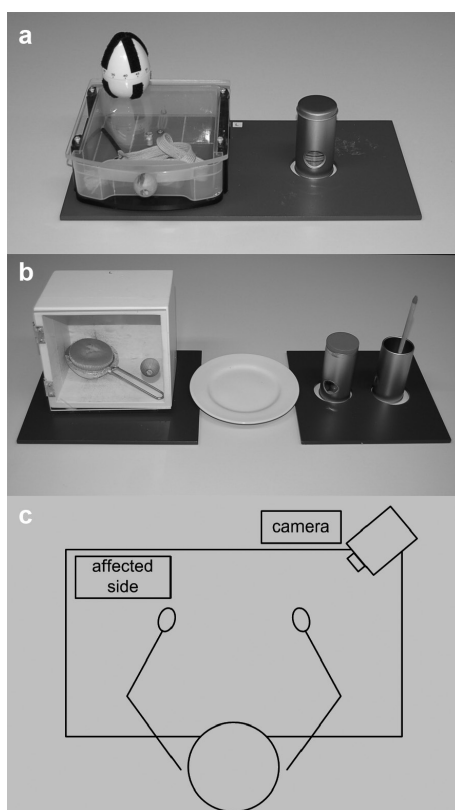
|                             | CP (n=25) | TDC (n=46) |
|-----------------------------|-----------|------------|
| <b>Age (y)</b>              |           |            |
| Mean (SD)                   | 4.7 (1.6) | 5.3 (1.4)  |
| Range                       | 2.7–8.0   | 2.5–8.0    |
| <b>Sex, n</b>               |           |            |
| Male                        | 10        | 23         |
| Female                      | 15        | 23         |
| <b>Affected side, n (%)</b> |           |            |
| Right                       | 10        | –          |
| Left                        | 15        | –          |
| <b>Dominant side, n (%)</b> |           |            |
| Right                       | –         | 41         |
| Left                        | –         | 5          |
| <b>MACS, n</b>              |           |            |
| I                           | 5         |            |
| II                          | 12        |            |
| III                         | 8         |            |

MACS, Manual Ability Classification System.



test instructor was one of three occupational therapists experienced in pediatric rehabilitation. A typically developing child was instructed by one of two occupational therapy students. All test instructors received a test manual and training to administer the tasks in a standardized manner. The test instructor was seated opposite the child and provided the instructions for each subtask, without indicating which hand had to be used. Before the child started, the test instructor demonstrated the tasks and checked whether the child had understood the instructions. The video camera was placed contralateral to the child's affected side (non-dominant side for the typically developing children) focused into the palm of the affected hand (Fig. 1c).

**Figure 1** The (a) beads task and (b) muffin task



The materials are positioned for a child with a left-sided paresis, as observed by the child. For a child with right-sided paresis, the setting of the materials is mirrored. The camera was positioned contralateral to the child's affected side (non-dominant side for the typically developing children) at a height of 2 meters focused into the palm of the affected hand (c).

## Scoring system

The video recordings were scored offline for the occurrence of grasping, holding, and releasing (i.e. motor behaviors) as well as the overall duration of use of the affected upper limb (non-dominant side in typically developing children). The average duration for scoring all the measurements of one child was 30 minutes. The frequency of the three behaviors was scored irrespective of the quality (e.g. grasping with the wrist in dorsal flexion or in palmar flexion were both scored). The participant could obtain maximally one point for each of the three behaviors during each subtask, resulting in a maximum frequency score of 48 (three behaviors  $\times$  four subtasks  $\times$  four repetitions). Thus, the frequency measure did not take into account whether a behavior was performed multiple times during one subtask. The observed total frequency was converted into a percentage of the maximum frequency. The frequency score during the 'demanding' beads task was termed the capacity score, whereas the frequency during the 'stimulating' muffin task was termed the performance score. When a child was unable to perform four repetitions of the task, the total frequency score was adjusted accordingly (e.g. when a child could perform only three repetitions, the maximal attainable frequency score was 36). In addition, the overall duration of use of the affected upper limb was scored for both the beads and the muffin task as a percentage of the total duration of each task. All motor behaviors related to the task performance contributed to the duration score, regardless of their success or quality. The difference in the duration of use between the beads task and the muffin task was defined as developmental disregard.

## Procedure

The children with CP were assessed twice by two occupational therapists with a time interval of approximately 2 weeks, as recommended by Terwee et al.<sup>17</sup> The first assessments of the children with CP were scored by both raters to determine the interrater reliability. In addition, the same rater scored the first assessments twice with at least 2 weeks in between to determine the intrarater reliability. The scorings of the first and second assessments of the children with CP by the same raters were used to determine the test-retest reliability. The assessment of the typically developing children was scored by one rater (the physical therapist), and was used together with the first assessment of the children with CP to determine the construct validity.

## Analysis

### Participants

The characteristics of the children with CP were compared with those of the typically developing children for age (two-sided independent *t*-test) and sex (Mann-Whitney *U* test).

**Table 2** Item statistics of 14-item MHC scale. Items are ordered from easy to difficult.

| Score  | Mean scores (SD) |             | Mann-Whitney U test<br><i>p</i> | Reliability (CP)        |                         |                          |             |
|--|------------------|-------------|---------------------------------|-------------------------|-------------------------|--------------------------|-------------|
|  | TDC (n=46)       | CP (n=25)   |                                 | Intraclass ICC (95% CI) | Interrater ICC (95% CI) | Test-retest ICC (95% CI) | SDD (%)     |
| Capacity (%)<br>(range 0–100)                | 98.1 (3.7)       | 76.6 (39.8) | 0.052                           | 1.00<br>(1.00–1.00)     | 0.98<br>(0.95–0.99)     | 0.98<br>(0.96–0.99)      | 5.1<br>14.0 |
| Performance (%)<br>(range 0–100)             | 100 (0.0)        | 54.8 (41.1) | <0.001                          | 1.00<br>(1.00–1.00)     | 0.99<br>(0.98–1.00)     | 0.99<br>(0.97–0.99)      | 4.5<br>12.5 |
| Developmental disregard (%)<br>(range 0–100) | 6.6 (5.3)        | 23.3 (15.0) | <0.001                          | 0.98<br>(0.96–0.99)     | 0.95<br>(0.90–0.98)     | 0.79<br>(0.57–0.90)      | 6.8<br>19.0 |

ICC, intraclass correlation coefficient; CI, confidence interval; SEM, standard error of measurement; SDD, smallest detectable difference.

### Validity

Construct validity was determined by comparing the scores of the children with CP with those of the typically developing children, based on the following hypotheses. Compared with the typically developing children, the children with CP were expected to score lower on capacity and performance, and higher on developmental disregard. Between-group differences (children with CP vs typically developing children) were tested with Mann–Whitney *U* tests and within-group differences (capacity vs performance in children with CP) with a Wilcoxon signed ranks test. Furthermore, the effects of sex and age on the three scores were examined by testing the differences between males and females using Mann–Whitney *U* tests and by correlating the three scores with age using Spearman's  $\rho$ . The mean score +2 SD of the typically developing children was used as a cut-off criterion to determine developmental disregard in individual children with CP.

### Reliability

The intrarater, interrater, and test–retest reliability of the capacity, performance, and developmental disregard scores were quantified with the intraclass correlation coefficient (ICC) and corresponding 95% confidence interval. An ICC greater than 0.70 was considered good.<sup>18</sup> A two-way random model for absolute agreement was used to distinguish between random variations and ‘real’ differences.<sup>18</sup> The standard error of measurement (SEM) was used to assess the absolute agreement between the first and second assessment, according to Bland and Altman.<sup>19</sup> The SEM was calculated using the within-subject SD ( $SEM = \sqrt{\text{error variance}}$ ). To determine the minimal change score in an individual that represented a real difference, the smallest detectable difference was calculated as  $1.96 \times \sqrt{2} \times SEM$ .<sup>19</sup>

## Results

### Participants

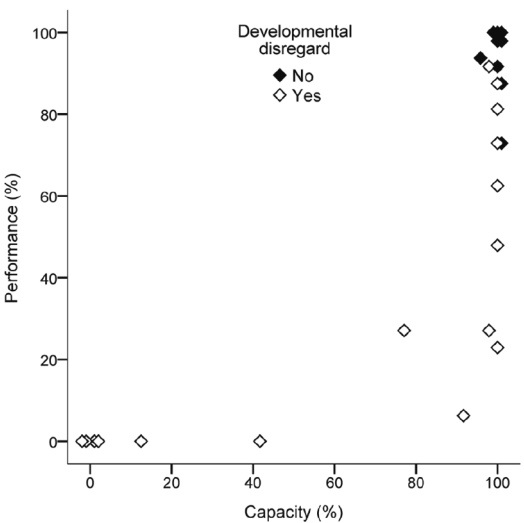
The characteristics of the children with CP and the typically developing children are presented in Table 1. The children with CP did not differ significantly from the typically developing children for sex ( $p=0.423$ ) or age ( $p=0.136$ ).

### Validity

Table 2 shows the capacity, performance, and developmental disregard scores for the children with CP and for the typically developing children. Seven of the children with CP could only perform two or three repetitions of the subtasks within 7 minutes. Consequently, their maximally attainable score on capacity and/or performance was adjusted to 24 and 36 respectively. The typically developing children scored almost maximally on capacity and performance, whereas the children with CP scored lower on capacity, almost reaching

statistical significance ( $p=0.052$ ), and significantly lower on performance ( $p<0.001$ ). Their performance scores were lower than their capacity scores ( $p<0.001$ ). Furthermore, children with CP scored three times higher on developmental disregard ( $p<0.001$ ). There were no effects of sex on the three scores for the children with CP ( $p>0.428$ ) and the typically developing children ( $p>0.095$ ), nor any effects of age on the performance and developmental disregard scores ( $p>0.248$ ). There was a small effect of age on the capacity score in the children with CP ( $\rho=0.436$ ;  $p<0.05$ ) and the typically developing children ( $\rho=0.758$ ;  $p<0.001$ ), indicating that older children performed better than younger children. The cut-off score for developmental disregard based on the mean scores +2 SD of the typically developing children was 17.2%. Based on this value, 64% of the children with CP could be identified as having developmental disregard. All individual scores are presented in Figure 2.

**Figure 2** Individual scores of children with CP ( $n=25$ ) on capacity (x-axis) and performance (y-axis)

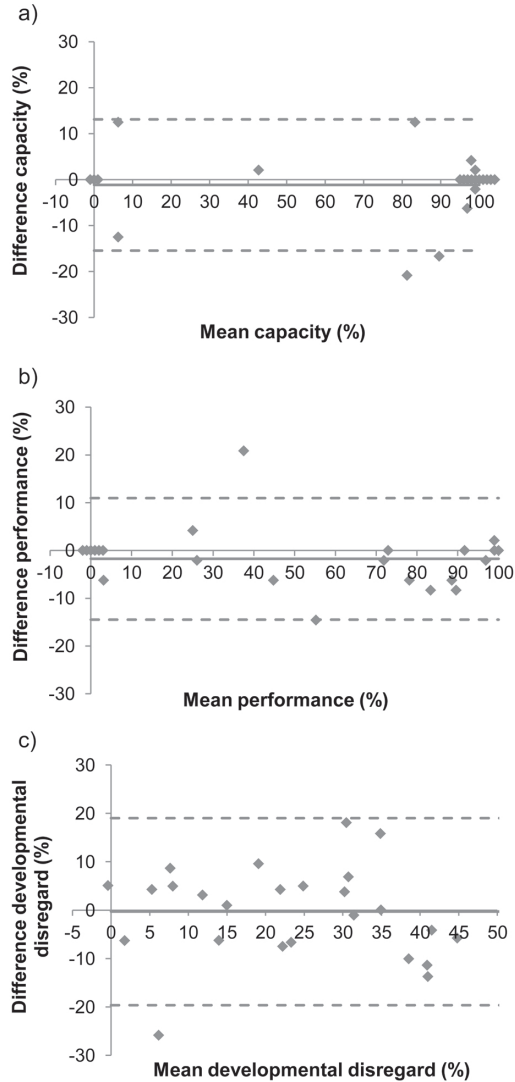


Individuals who were identified as having developmental disregard (i.e. a developmental disregard score  $>17.2\%$ ;  $n=16$ ) are depicted by white diamonds, whereas children without developmental disregard are depicted by black diamonds.

### Reliability

The intra- and interrater reliability of the capacity, performance, and developmental disregard scores were excellent, with ICCs ranging from 0.95 to 1.00 (Table 2). The test-retest reliability was excellent for the capacity and performance scores, whereas it was good for the developmental disregard score (ICCs ranged from 0.79 to 0.99). The mean

**Figure 3** The absolute agreement between two repeated assessments of the same rater in children with CP ( $n=25$ ) according to Bland and Altman.<sup>19</sup>



The difference score between the two assessments is plotted against the mean score for capacity (a), performance (b), and developmental disregard (c). The solid line represents the mean difference, the dotted lines represent the limits of agreement.

differences between the first and second assessments in children with CP were  $-1.2\%$  (SD 7.1) for capacity,  $-1.8\%$  (SD 6.4) for performance, and  $-0.3\%$  (SD 9.7) for developmental disregard. The absolute agreement between the two assessments is presented in Figure 3. The SEMs ranged from 4.5 to 6.8%, which resulted in smallest detectable differences of between 12.5% and 19.0%.

## Discussion

The results of this study indicate that the three scores of the VOAA-DDD-R (i.e. capacity, performance, and developmental disregard) are both valid and reliable. The construct validity was determined by comparing the scores of children with CP with those of typically developing children, because there is no criterion standard available in the literature for the frequency and duration of use of the affected upper limb during bimanual activities. Children with CP had lower scores than typically developing children for capacity (77 vs 98% respectively) and significantly lower scores for performance (55 vs 100% respectively), yielding much higher scores for developmental disregard (23 vs 7% respectively). In addition, the variability in the CP group was much higher compared with the typically developing children. Furthermore, the older children performed better than the younger ones on the capacity score, which may have been related to improvements in bimanual performance that are related to development. This finding needs to be taken into account by therapists when assessing younger children with the VOAA-DDD-R.

Based on the cut-off score for developmental disregard of typically developing children (i.e. 17%), 64% of the children with CP could be identified as having developmental disregard (Fig. 2). This cut-off value is close to the cut-off value reported in our previous study on the VOAA-DDD (14%).<sup>13</sup> These results confirm our hypothesis that many children with CP show a discrepancy between what they can do with their affected upper limb when bimanual activity is demanded (i.e. capacity) and what they actually do when bimanual activity is merely stimulated (i.e. performance). These test scores can be used as a basis for designing an individually tailored rehabilitation intervention.<sup>15</sup> For instance, Figure 2 shows that the six children with a low capacity score (0–40%) scored 0% on performance. Based on these scores it is advisable that these children are primarily trained to improve their upper-limb capacity. Remarkably, even eight children with a (near) maximum capacity score of 100% showed some degree of developmental disregard, whereas nine others with a maximum capacity did not. This pattern of results suggests that a (nearly) optimal capacity is needed to prevent the occurrence of developmental disregard, but that such a score certainly provides no guarantee for the absence of developmental disregard. Thus, these children should all be carefully monitored for signs of developmental disregard and offered appropriate training, e.g. constraint-induced movement therapy. On the other hand, one or two children with a somewhat lower

performance than their optimal capacity scores did not seem to have developmental disregard based on the duration of use of their affected upper limb.

The VOAA-DDD-R showed excellent intra- and interrater reliability, as indicated by high ICCs for capacity, performance, and developmental disregard. Reliability in this context means that repeated measurements result in similar outcomes,<sup>17,18</sup> which are not influenced by characteristics of the instrument, differences in performance by the same rater, differences between multiple raters, or by the natural variability within a subject. The ICC values in the present study indicated that the repeated scoring of the assessments was very stable both within (intrarater reliability) and between raters (interrater reliability). This suggests that when a child is assessed twice by the same rater or by two different raters, the results are generally the same and not affected by the measurement instrument. The test-retest reliability was excellent for the capacity and performance scores and good for developmental disregard. Thus, the variability between two assessments caused by variation of the child's behavior was larger than by variation caused by the raters. In addition, the results indicate that with repeated testing the frequency scores were more stable than the duration scores. This can be explained by the fact that for the frequency scores a child could obtain maximally one point for each behavior per subtask, which renders the frequency scores more stable but also less sensitive to repeated behaviors within a subtask. Nevertheless, the absolute agreement between the repeated assessments was good, as indicated by SEMs between 4.5 and 6.8%. These results imply that, when two groups of children with CP are compared, a group difference of 5.1% on capacity, 4.5% on performance, and 6.8% on developmental disregard can be regarded as a real difference and not due to natural variation. For individual children, a change in the VOAA-DDD-R scores needs to be larger to be significantly different, because the smallest detectable differences ranged from 12.5 to 19.0%. These results indicate that although the VOAA-DDD-R is suitable to detect differences between groups, it needs to be further refined to be able to detect smaller changes in individual children.<sup>19</sup>

Until now, no reliable and valid measure of developmental disregard has been available in the literature. In this perspective, the VOAA-DDD-R is a valuable addition to the existing measures of affected upper-limb use in children with CP. Because the VOAA-DDD-R consists of common daily-life tasks that are attractive and meaningful for all children, it may also have merits for other groups of children with unilateral upper-limb disability, for instance children with peripheral nerve damage, traumatic brain injury, or stroke. A limitation of the present study is that the responsiveness (i.e. sensitivity to change) was not investigated. Thus, future studies need to examine the responsiveness of the VOAA-DDD-R to determine its usefulness and sensitivity in intervention studies. Another limitation is that one could argue that the VOAA-DDD-R is not truly a test of upper-limb performance in daily life, because it requires a standardized test situation. Yet, a drawback of real-life assessments is that they may be too subjective. For instance, self-report questionnaires<sup>20,21</sup> are usually completed by the child's parents or caregivers



with a great influence of personal perspectives and proneness to inconsistencies. Recent developments in the use of wearable wrist activity monitors to assess actual daily-life use of the affected upper limb are promising,<sup>22</sup> but such monitors have only been tested during standardized activities as well. Finally, the construct validity was determined based on the assessment of typically developing children, who are expected to have no limitations in capacity and performance and show no developmental disregard. To confirm that the cut-off value for developmental disregard used in this study was indeed valid, we need to investigate other groups of children with CP with and without developmental disregard as determined by, for example, experts.

In conclusion, this study showed that the VOAA-DDD-R, using a simplified scoring system, is equally reliable, when performed 2 weeks apart, and as valid as the original VOAA-DDD when applied by trained occupational and physical therapists to children with unilateral spastic CP (2y 6mo to 8y). By comparing the use of the affected upper limb during a task demanding the use of both hands compared with a task merely stimulating bimanual activity, upper-limb capacity, performance, and developmental disregard can reliably and validly be assessed offline with a computer-supported video scoring system.

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# 7

## Development and psychometric properties of the Hand-Use-at-Home questionnaire to assess amount of affected hand use in children with unilateral paresis

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## Abstract

To describe the development of the parent-rated Hand-Use-at-Home questionnaire (HUH) that assesses the amount of spontaneous use of the affected hand in children with unilateral paresis, and to test its internal structure, unidimensionality and validity.

Parents of children with Unilateral Cerebral Palsy (UCP) and professionals participated in the development of the HUH. To examine internal validity, data of 322 children (mean age 6y 7mo, range 3-10y) with UCP (n=131) or Neonatal Brachial Plexus Palsy (NBPP) (n=191) were collected. Rasch analysis was used to examine discriminative capacity of the 5-point rating scale as well as unidimensionality and hierarchy of the item set. Additionally, data of 55 typically developing children (TD) (6y 9mo, SD 2y 5mo) were used to examine construct validity.

The 5-point rating scale was disordered in all items and was collapsed to form a well-structured 3-point rating scale. Ten misfitting or redundant items were removed. Eighteen hierarchically ordered bimanual items fitted a unidimensional model within acceptable range. The HUH significantly discriminated between the three groups (TD,NBPP,UCP):  $H(2)=118.985, p<0.001$  supporting its construct validity.

The Hand-use-at-Home questionnaire is a valid instrument to assess the amount of spontaneous use of the affected hand in children with unilateral upper limb paresis.

Unilateral cerebral palsy (UCP) and neonatal brachial plexus palsy (NBPP) constitute two of the most common forms of unilateral non-progressive neurological conditions in children. In both UCP and NBPP, repeated failure to perform activities with the affected arm and hand may lead to decreased spontaneous use. Consequently, children with a unilateral paresis often use their affected upper limb less frequently in bimanual activities than might be expected based on their functional capacities.<sup>1-4</sup> This discrepancy between capacity and performance<sup>5</sup> (referred to as developmental disregard) has been reported in UCP<sup>6, 7</sup> and NBPP<sup>8</sup> and may lead to a vicious circle of decreasing upper-limb use. Despite intensive rehabilitation, parents often report that their children seldom spontaneously use the affected arm and hand at home.<sup>2</sup> This underuse results in an inefficient task performance and subsequently, may hamper social participation.<sup>9</sup> Clinicians are challenged to identify differences in upper-limb use between the therapeutic setting and the home situation. Spontaneous hand-use reflects automatized upper-limb motor control, but currently there is a lack of instruments assessing the amount of spontaneous use in daily life. Clinical tests focus on effective use of the affected upper limb (Assisting Hand Assessment<sup>10, 11</sup>) or on developmental disregard (Video Observation Aarts and Aarts module Determine Developmental Disregard Revised<sup>4</sup>), but are actually clinical surrogates for daily life performance that is influenced by personal and environmental factors.<sup>5</sup> A parent-reported measure of the amount of hand-use during every-day activities may be a valuable addition to the existing clinical assessments. Valid and frequently used parent-rated questionnaires measure various aspects of actual daily life performance, but none of them assesses the amount of affected hand-use. The ABILHAND-Kids<sup>12</sup> assesses manual ability based on unimanual and bimanual activities, whereas the Children's Hand-use Experience Questionnaire<sup>9</sup> focuses on a child's perspective of affected hand-use in bimanual activities. Currently, the only parent-completed measure of the amount of affected upper limb use at home is the Pediatric Motor Activity Log Revised (PMAL-R).<sup>13</sup> The PMAL-R evaluates the quality ('how well') and frequency ('how often') of affected upper limb use. However, psychometric properties of the 'how often' subscale have not yet been evaluated<sup>13</sup> and the majority of the PMAL-R items (16/22) are unimanual,<sup>14</sup> even though most daily activities are bimanual by nature. Although parent-reported measures are subjective, more objective measures (e.g. accelerometry, video-observation) cannot readily be used in young children,<sup>15</sup> are time-consuming or influence daily-life behavior. Against this background, we developed the parent-reported Hand-Use-at-Home questionnaire (HUH) to evaluate how often the affected upper limb is spontaneously used in typical daily life activities of children with unilateral upper limb paresis, aged three to ten years. In this age group play and self-care activities can easily be observed by parents.

In this paper we describe the development of the HUH. Content validity was established by means of a consensus process among professionals and parents. We used Rasch analysis to examine the appropriateness of the items' rating scale and to test whether the items together form a unidimensional and hierarchical construct. Appropriateness of

the rating scale refers to whether or not the parents' ratings on the items can be differentiated as clearly as the points of the rating scale allow. For the assessment of construct validity, we examined if the amount of spontaneous hand-use in children with UCP or NBPP would differ significantly from each other as well as from typically developing (TD) children of the same age. We hypothesized that children with UCP or with NBPP obtain lower scores on the HUH compared to TD children and that children with UCP would have a lower score for spontaneous use of the affected hand than children with NBPP.

## **Method**

### **Development of the HUH**

To validly assess the amount of spontaneous hand-use, the HUH needed to consist of activities that (1) are executed at home, (2) are typical of the daily lives of children aged three to ten years, (3) can be observed by parents, and (4) are suitable to test the degree to which the affected arm and hand are used. Activities can be ranked according to the degree to which they elicit a bimanual approach, starting with activities that can easily be performed in consecutive unimanual steps (e.g. fill a glass with water from a tap) up to activities that require the simultaneous use of both hands (e.g. closing a zipper). However, how these activities are ranked in children with unilateral paresis is unknown.

The first version of the HUH consisted of a broad range of age-related activities derived from interviews with parents and therapists. Subsequently, parents of 20 children with UCP rated for each activity (1) the frequency of spontaneous use of the affected hand on a 5-category rating scale (never/seldom/sometimes/mostly/always), (2) the importance of the activity on a 3-category rating scale (not at all/somewhat/very important), (3) whether or not it was applicable to their child (never occurs/not applicable due to age), and provided additional comments. Next, an expert-panel of six experienced pediatric occupational therapists determined whether activities (i.e. play, self-care) were relevant for children aged three to ten years and categorized activities as either 'unimanual' or 'bimanual'. For bimanual tasks, it was determined if activities demanded the use of the affected hand or if they could also be executed unimanually in consecutive steps, as both kinds of activities were deemed necessary in the HUH. In addition, the experts could suggest additional activities and comment on the phrasing of the questions. This yielded a list consisting of 31 activities. A new group of 20 parents of children with UCP completed this 31-item version of the HUH and commented on the questionnaire. This resulted in the deletion of the three activities they rated as 'not important at all'. This version, consisting of 28 questions (22 bimanual and 6 unimanual activities), was subsequently reviewed by another small group of five parents, who expressed only one minor concern in the wording. Thus, the final version of the HUH had 28 items, for which frequency of spontaneous use of the affected hand was rated on a 5-category rating scale: (almost)

**Table 1** Demographic and clinical characteristics of the children

|                               | UCP<br>n=131 | NBPP<br>n=191 | TD<br>n=55  | p-value (group<br>differences) |
|-------------------------------|--------------|---------------|-------------|--------------------------------|
| Mean age, <i>years;months</i> | 6;6          | 6;9           | 6;9         | 0.59 <sup>a</sup>              |
| SD                            | 2;2          | 2;0           | 2;5         |                                |
| Range                         | 3;0 – 10;9   | 3;0 – 10;5    | 3;5 – 10;11 |                                |
| Gender, <i>n</i>              |              |               |             | 0.726 <sup>b</sup>             |
| Male/Female                   | 63/ 68       | 95/ 96        | 24/ 31      |                                |
| Affected side, <i>n</i>       |              |               |             | 0.06 <sup>b</sup>              |
| Right/ Left                   | 75/ 56       | 89/ 102       | NA          |                                |
| MACS*, <i>n (%)</i>           |              |               |             |                                |
| I                             | 31 (23.7)    | NA            | NA          |                                |
| II                            | 52 (41.2)    |               |             |                                |
| III                           | 41 (35.1)    |               |             |                                |
| lesion extent*, <i>n (%)</i>  |              |               |             |                                |
| C5                            |              | 2 (1.0)       |             |                                |
| C5-C6                         |              | 120 (62.8)    |             |                                |
| C5-C7                         | NA           | 35 (18.3)     | NA          |                                |
| C5-C8                         |              | 15 (7.9)      |             |                                |
| C5-T1                         |              | 17 (8.9)      |             |                                |
| not specified                 |              | 2 (1.0)       |             |                                |

\*extracted from the medical record, UCP= Unilateral Cerebral Palsy, NBPP= Neonatal Brachial Plexus Paresis, TD= Typically Developing, MACS= Manual Ability Classification System, NA= not applicable, <sup>a</sup>One way ANOVA,

<sup>b</sup>Pearson Chi-Square

never, sometimes, regularly, often and (almost) always. Seven items had a not applicable option to reduce possible bias related to age (e.g. 'buttering bread') or gender ('dress doll or teddy bear') (Table 2). For the present study, the HUH was available online but, if preferred, a paper version was available as well.

## Participants and design

Three groups of parents of Dutch children aged three to ten years participated: parents of children with UCP or NBPP and parents of TD children. For the UCP group, 11 of the 13 pediatric rehabilitation centers that are part of the Dutch Collaboration for Implementation of the Pirate Concept (LIPIC)<sup>16</sup> recruited 131 parents who completed the questionnaire. The NBPP group consisted of 192 parents who completed the HUH as part of a cross-sectional study in children and adolescents with NBPP, that was conducted at the Leiden University Medical Centre. The TD group consisted of 55 parents who were recruited from the personnel of one rehabilitation center (Sint Maartenskliniek) and their families, friends and neighbors. They completed a version of the HUH in which the term 'affected hand' was replaced by 'non-preferred hand'. All participants gave written informed consent.



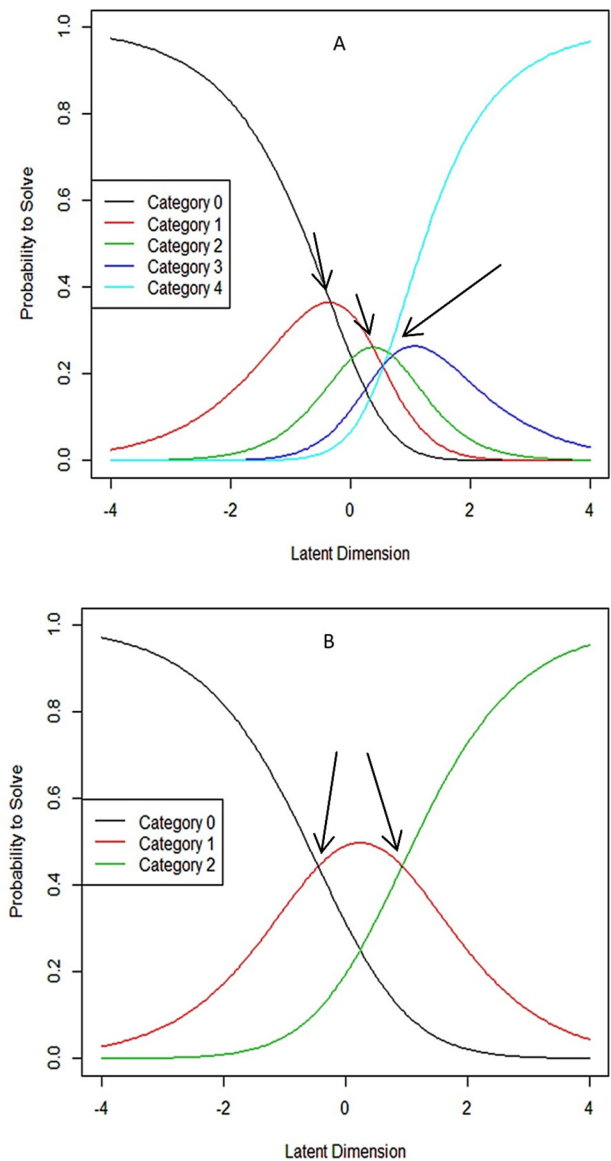
**Table 2** HUH-items with item measure estimates and fit statistics

| Items HUH<br>How often does your child use the affected hand spontaneously to<br>... | Bimanual (B)<br>Unimanual (U) | Scored<br>not-applicable<br>(%) |
|--|-------------------------------|---------------------------------|
| pick up a large ball (football size) from the floor?                                 | B                             |                                 |
| catch a large ball (football size) while playing ball games?                         | B                             |                                 |
| move a chair?  | B                             |                                 |
| hold a book while reading or looking at pictures?                                    | B                             |                                 |
| throw a large ball (football size) while playing ball games?                         | B                             |                                 |
| assist in closing a zipper (jacket/cardigan/pants etc.)?                             | B                             |                                 |
| play with construction toys (Lego/Duplo/etc.)?                                       | B                             |                                 |
| stabilise paper while drawing or writing?  | B                             |                                 |
| assist undressing the upper body (remove singlet, shirt, sweater)?                   | B                             |                                 |
| hold a toy while playing on the floor?   | B                             |                                 |
| assist with tearing off and folding toilet paper when using the toilet?              | B                             | 4.7                             |
| assist with opening, closing buttons (jacket/blouse/pants etc.)?                     | B                             | 6.5                             |
| hold a shovel while digging a hole in the sand (beach/garden/sandbox)?               | B                             |                                 |
| assist putting toothpaste on a toothbrush?   | B                             | 9.3                             |
| assist with buttering bread?   | B                             | 11.5                            |
| pull off socks?  | B                             |                                 |
| hold a toy while walking?  | B                             |                                 |
| hold a fork while cutting food?  | B                             | 14.9                            |
| * assist dressing the upper body (put on singlet, shirt, sweater)?                   | B                             |                                 |
| * hold a toy while playing at a table?   | B                             |                                 |
| * assist (un)dressing a doll or teddy bear?  | B                             | 37.0                            |
| * tie shoelaces?   | B                             | 36.3                            |
| * wave at somebody?  | U                             |                                 |
| * open/close the tap?  | U                             |                                 |
| * stroke a pet or cuddle toy?  | U                             |                                 |
| * open/close a door?   | U                             |                                 |
| * push buttons (radio/TV/light etc.)?  | U                             |                                 |
| * close a drawer or cupboard?  | U                             |                                 |

Included HUH items are ordered based on increasing difficulty, \* item excluded from final HUH scale, # item location is the mean item-measure i.e. the mean of item-measures 1 and 2, MSQ= mean square value, TD= typically developing, #Scores of TD children were not used to establish the final HUH scale

| Item location # | Item-measure 1 | Item-measure 2 | MSQ infit | Score 0 in TD group (%) | Score 1 in TD group (%) | Score 2 in TD group (%) |
|-----------------|----------------|----------------|-----------|-------------------------|-------------------------|-------------------------|
| -1.07           | -2.24          | 0.10           | 0.92      | 0                       | 0                       | 100                     |
| -0.57           | -1.71          | 0.57           | 0.94      | 0                       | 0                       | 100                     |
| -0.45           | -1.83          | 0.93           | 0.81      | 5.5                     | 21.8                    | 72.7                    |
| -0.12           | -1.39          | 1.15           | 0.78      | 5.5                     | 10.9                    | 83.6                    |
| -0.12           | -1.39          | 1.15           | 1.08      | 18.2                    | 9.1                     | 72.7                    |
| -0.01           | -0.81          | 0.79           | 0.96      | 1.8                     | 3.6                     | 94.5                    |
| 0.04            | -1.27          | 1.36           | 0.80      | 3.6                     | 10.9                    | 85.5                    |
| 0.11            | -1.23          | 1.45           | 1.21      | 3.6                     | 34.5                    | 61.8                    |
| 0.24            | -0.45          | 0.93           | 0.91      | 0                       | 1.8                     | 98.2                    |
| 0.27            | -1.41          | 1.94           | 0.90      | 14.5                    | 36.4                    | 49.1                    |
| 0.53            | 0.13           | 0.93           | 0.80      | 9.1                     | 9.1                     | 81.9                    |
| 0.54            | -0.27          | 1.34           | 0.88      | 1.8                     | 0                       | 98.2                    |
| 0.59            | 0.02           | 1.16           | 0.78      | 3.6                     | 1.8                     | 94.5                    |
| 0.76            | 0.36           | 1.15           | 1.35      | 18.2                    | 5.5                     | 76.4                    |
| 0.82            | -0.04          | 1.68           | 1.09      | 25.5                    | 3.6                     | 70.9                    |
| 0.96            | 0.65           | 1.27           | 0.88      | 23.6                    | 23.6                    | 52.7                    |
| 1.14            | -0.19          | 2.47           | 1.13      | 40                      | 40                      | 20                      |
| 2.15            | 1.45           | 2.86           | 1.39      | 23.6                    | 3.6                     | 72.7                    |

**Figure 1** Item Characteristic Curves plotted (for item 6) as an example for (dis)ordered rating scales: before (**A**, disordered rating scale) and after collapsing item categories (**B**, ordered rating scale)



Data were collected between October 2013 and February 2015 and the study was approved by the medical ethical committee Arnhem-Nijmegen (2013/395), the medical-ethics committee of the Leiden University Medical Centre (P14.071), as well as by the executive boards of the participating institutions.

### Statistical analysis

Missing values in the questionnaires were replaced with predicted values using the Expectation-Maximization technique in SPSS (IBM SPSS Statistics 20.0). A not applicable score was recorded as 0 (never).

We used Rasch analysis (the partial credit model for polytomous rating scales) to examine the appropriateness of the rating scales and to identify misfitting items. With Rasch analysis, item-measures and person-measures are estimated from the response patterns and are expressed on a common log-odds unit scale. Rasch rating scale analysis<sup>17</sup> was performed to examine appropriateness of the item rating scale to check whether item threshold values were ordered (i.e. increasing threshold values for higher category scores). In case of non-ordered, reversed, thresholds item categories were collapsed until ordered threshold values appeared. To determine whether item scores could be summed, the data were tested for unidimensionality using mean square infit statistics. Values between 0.71 – 1.4 are considered as an acceptable fit to the Rasch model's expectation.<sup>18, 19</sup> A person-item map was plotted to examine the distribution of item- and person-measures to allow identification of levels of spontaneous bimanual hand-use that are poorly assessed by the items. Rasch analysis was performed using the R package eRm.<sup>20</sup>

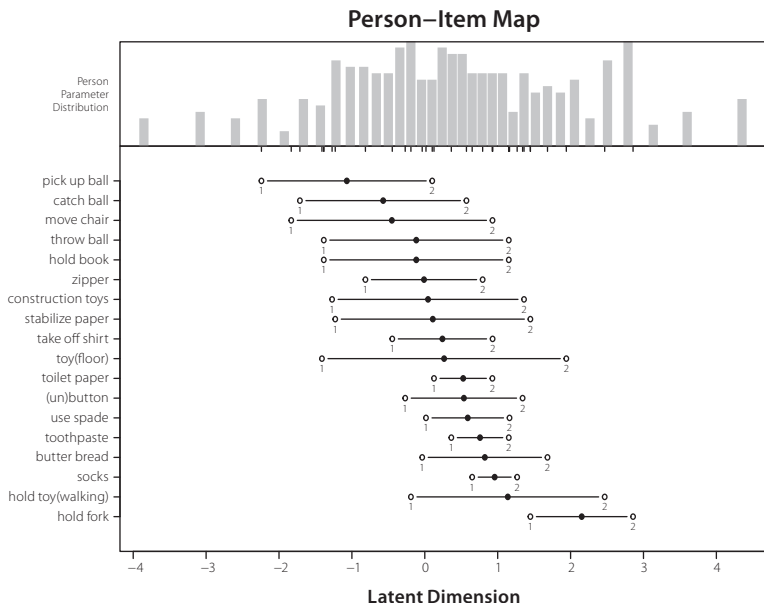
Internal consistency of the HUH was examined by calculating Cronbach's  $\alpha$ . An  $\alpha$  between 0.7 and 0.9 was considered adequate.<sup>21</sup>

Construct validity was examined by testing the differences in mean HUH sum scores between children with UCP, children with NBPP and TD children using Kruskal Wallance-H tests with pair-wise comparisons using the Dunn-Bonferroni approach. We also correlated the individual HUH sum scores with age, affected side and gender to examine whether they were influenced by these characteristics using Pearson and point-biserial correlation, respectively.

## Results

A total of 378 questionnaires were returned. One questionnaire of the NBPP group was discarded because more than half of the scores were missing. Nine questionnaires had one to three missing values and were used for analysis after imputation of missing values. Thus, for Rasch analysis, 322 questionnaires from the UCP (n=131) and NBPP (n=191) groups were available. The TD group was not included in the Rasch analysis. The characteristics of all children are presented in Table 1. There were no significant differences in age or gender between the three groups.

**Figure 2** Person-Item map; distribution of HUH-person-measures and item-measures



Every vertical bar of the person parameter distribution represents a raw total score. The bars for the lowest and highest total scores (0 and 36) are omitted from the figure. Item-measures for scores 1 and 2 for each item are represented by the small stripes below the bars and by the corresponding open dots. Black dots indicate the mean item-measure for each item (item location). Note: the latent dimension on the X-axis is expressed in logits.

When the data fit the Rasch model, item thresholds (i.e., boundaries between response categories indicated with arrows) should monotonically increase. In part A the thresholds of the item characteristic curves are disordered and there is discordance between the category probabilities. Category 3 seems to be redundant. In this case a child with a person-measure 0.7 has equal probabilities for a category-score 2, 3 or 4 (regularly, often, always) for spontaneous hand-use. (B): After collapsing item categories, the thresholds are ordered. A child with a person-measure between -0.4 and 0.9 logits has the largest probability to obtain a score 1 (regularly, often). With a person-measure > 0.9 logits, the probability of a category 2 score (always) increases rapidly.

### Establishing the final HUH item set

Two items ('dress a doll or bear' and 'tie shoelaces') with more than 35% 'not applicable' responses were excluded. Rasch analysis showed that the 5-point rating scales were

disordered for most items (see Figure 1 for explanation) and that some response options were underused. Thus, the rating scales were collapsed into three categories (i.e., never + sometimes = score 0; regularly + often = score 1; always = score 2), which resulted in ordered category thresholds for all items, except for the unimanual items. For these unimanual items, the scales were disordered to the extent that only dichotomizing was possible. Therefore, these items were deleted. Item 'play at the table' correlated strongly ( $r=0.92$ ) with 'play on the floor' and showed strong inter-item correlations ( $r>0.74$ ) with two other items. Therefore, it was considered redundant<sup>21</sup> and omitted. Item 'dress upper body' was removed as it correlated strongly with 'undress upper body' ( $r=0.76$ ). Thus, 18 items fitted in the final scale with total scores ranging from 0 to 36 points. Item infit statistics (Table 2) were all within the acceptable range indicating good fit to the Rasch model. The highest mean square value was for 'hold fork, cutting' and indicated that, although within the acceptable range, data from this item were less predictable than the model expected.

### Psychometric properties

Table 2 presents the item-measure estimates for scores 1 and 2 after collapsing the response categories into a 3-point item-score. The lowest item-measure indicates score 1 (i.e., the degree to which the activity elicits regular or often use of the affected hand) and the highest score 2 (i.e., the degree to which the activity always elicits use of the affected hand). Item-measures ranged from -2.24 to +2.86 logits. Two items ('throw a ball' and 'hold a book') had equal item-measures; as they each represent a completely different kind of activity (outdoor/gross motor play vs. indoor/seated activity) both were kept in the scale.

To obtain the HUH-score (person-measure) the 3-point item-scores were summed and subsequently converted into a logit score (Table 3). Figure 2 shows the distribution of HUH person-measures and item-measures for children with unilateral paresis ( $N=322$ ). The person-measure estimates reflect the extent to which a child spontaneously uses the affected hand. Person-measures ranged from -4.69 to +5.17 logits and were not normally distributed in the sample. Thirteen children had a person-measure below the lowest item-measure of -2.24 logits (0–3 raw sum score). Thirty-five children had person-measures above the highest item-measure of 2.86 logits (scores 33–36). The three children with a zero sum score (-4.69 logits) were all diagnosed with UCP and scored seldom or never, even on the easiest items. The highest HUH-score of 5.17 logits (36 points) was obtained by 7% of the sample (23 children with NBPP) who always spontaneously used their affected hand in all 18 activities. The lowest item-measure was for 'pick up a large ball', which is the most provoking activity to use the affected hand regularly or often (achieved by 91.3% of the sample), and 'holding a fork while cutting food' was the most difficult item to always elicit the use of the affected hand (achieved by 15% of our sample).

Internal consistency (Cronbach's  $\alpha$ ) of the 18-item HUH was 0.941. HUH-scores showed weak correlations with affected side ( $r_{pb}=0.127$ ,  $p=0.023$ ), gender ( $r_{pb}=0.109$ ,  $p=0.051$ )

**Table 3** Conversion table of sum scores to obtain the HUH-score (person-measure) in logits

| Sum score | HUH-score (logits) | SE   | Sum score (continued) | HUH-score (logits) | SE   |
|-----------|--------------------|------|-----------------------|--------------------|------|
| 0         | -4.695             | NA   | 19                    | 0.513              | 0.37 |
| 1         | -3.853             | 1.04 | 20                    | 0.649              | 0.37 |
| 2         | -3.082             | 0.76 | 21                    | 0.786              | 0.37 |
| 3         | -2.597             | 0.64 | 22                    | 0.925              | 0.37 |
| 4         | -2.230             | 0.57 | 23                    | 1.066              | 0.38 |
| 5         | -1.927             | 0.53 | 24                    | 1.211              | 0.38 |
| 6         | -1.166             | 0.50 | 25                    | 1.361              | 0.39 |
| 7         | -1.431             | 0.47 | 26                    | 1.518              | 0.40 |
| 8         | -1.218             | 0.45 | 27                    | 1.684              | 0.41 |
| 9         | -1.022             | 0.44 | 28                    | 1.861              | 0.43 |
| 10        | -0.838             | 0.42 | 29                    | 2.053              | 0.45 |
| 11        | -0.665             | 0.41 | 30                    | 2.265              | 0.47 |
| 12        | -0.500             | 0.40 | 31                    | 2.506              | 0.51 |
| 13        | -0.343             | 0.39 | 32                    | 2.788              | 0.56 |
| 14        | -0.191             | 0.39 | 33                    | 3.134              | 0.62 |
| 15        | -0.044             | 0.38 | 34                    | 3.599              | 0.75 |
| 16        | 0.098              | 0.38 | 35                    | 4.352              | 1.03 |
| 17        | 0.238              | 0.37 | 36                    | 5.174              | NA   |
| 18        | 0.376              | 0.37 |                       |                    |      |

HUH-score= Hand-Use-at-Home score, SE= standard error, NA= not applicable

After recalculating parents' ratings for the 18 activities from 5 categories into a 3 points-score (i.e., *never/sometimes* = score 0; *regularly/often* = score 1; *always* = score 2) the scores can be summed. The (raw) sum score can be converted into a person-measure for spontaneous hand-use (HUH-score), expressed in logits.

and age ( $r_p=0.149$ ,  $p=0.007$ ). Girls and left-side affected children had slightly higher HUH-scores and scores increased with age. Equal trends for age and gender were found in TD children. Children with a maximum HUH-score were aged between 4y;5mo and 9y;7mo.

### Construct validity

Scores for the amount of spontaneous hand-use differed between groups ( $H(2)=118.985$ ,  $p<0.001$ ). Post-hoc tests showed that all group differences were significant ( $p<0.001$ ). The TD group had the highest amount of spontaneous hand-use (median HUH-score: 2.51 logits, range 0.65-5.17) with 5.5% of the children obtaining the maximum score. The lowest median HUH-score was for the UCP group (-0.343 logits, range -4.70-2.79). The NBPP group had an intermediate median HUH-score of 1.07 logits (range -3.08-5.17).

## Discussion

Although underuse of the affected limb in children with unilateral paresis is recognized as an important problem<sup>7,22</sup>, there is no valid and reliable measure to assess daily-life amount of hand-use. We developed the parent-rated Hand-Use-at-Home questionnaire (HUH) to assess the amount of spontaneous hand-use at home in three to ten year old children. We examined its internal structure, unidimensionality and construct validity. The results showed that 18 hierarchically ordered bimanual activities fitted the unidimensional Rasch model after collapsing the rating scale with 5 response options into a 3-point item-score. HUH-scores clearly discriminated between children with UCP or NBPP and TD children, supporting its construct validity. Compared to the constructs of the CHEQ (perceived quality of performance) and ABILHAND-Kids (manual ability), the HUH adds an important aspect of upper-limb performance: the amount of spontaneous hand-use. Applying the HUH will provide a good opportunity for a dialogue between therapists and parents about parents' observations and expectations and about the possibilities to enhance spontaneous hand-use in the home environment.

The PMAL-R is the only measure with a subscale for amount of hand-use, but the over-representation of unimanual items in this interview-based assessment does not reflect the large proportion of bimanual activities that children encounter in daily life. In our study we examined six unimanual items (Table 2) and found that some categories of the 5-point rating scale were used infrequently or not at all. No adequate re-ordering of the rating scales could be established for these items. For most unilateral items, the frequencies for score 0 (never, sometimes) in TD children were very high (60-80%). Altogether, we concluded that unimanual activities are not suitable to measure spontaneous use of the affected hand in our target groups.

The spread of the item-measures (Figure 2) indicates that the activities were adequately eliciting spontaneous hand-use ranging from 'easy' to 'difficult'. The HUH-scores (person-measures) had a much wider range, which could indicate floor and ceiling effects, although effects smaller than 15% are usually considered acceptable.<sup>23</sup> With only 4% of our sample reaching a sum score beneath -2.24 logits, the floor effect is negligible. Yet, 9.2% of the children scored above 2.86 logits and these children (all NBPP) always used their affected hand in at least 15 of the 18 HUH items. It indicates that extra items might be needed to differentiate in the upper part of the scale for children with NBPP, although in this group normal hand-use may reflect spontaneous recovery occurring in about 70% of the cases.<sup>24</sup>

Our results showed that the HUH sum score discriminates between TD children and children with unilateral upper limb paresis. Although the results confirmed that TD children had the highest scores, only 3 children (5.5%) had a maximum score indicating that TD children did not always use both hands in all bimanual activities either. Moreover, in only six activities the score always was attained by 95% of the TD children (Table 2). This suggests that not every bimanual activity is always provoking bimanual task execution,



not even in children that are able to use both hands simultaneously and well-coordinated. Therefore, we suggest that 100% bimanual task execution in children with UCP or NBPP cannot be expected.

The HUH was designed as a measure of spontaneous hand-use from a parental perspective. Parents can be considered as experts of the abilities and needs of their children,<sup>25</sup> especially in young children, and are therefore a valid source of information. It has to be acknowledged, however, that the comparison between TD children and children with UCP or NBPP might be influenced by the fact that parents of the TD group needed to focus on the non-preferred hand during bimanual activities. Because these parents were not used to observe their child's use of the non-preferred hand, it often took them substantially more time to observe their child and complete the HUH.

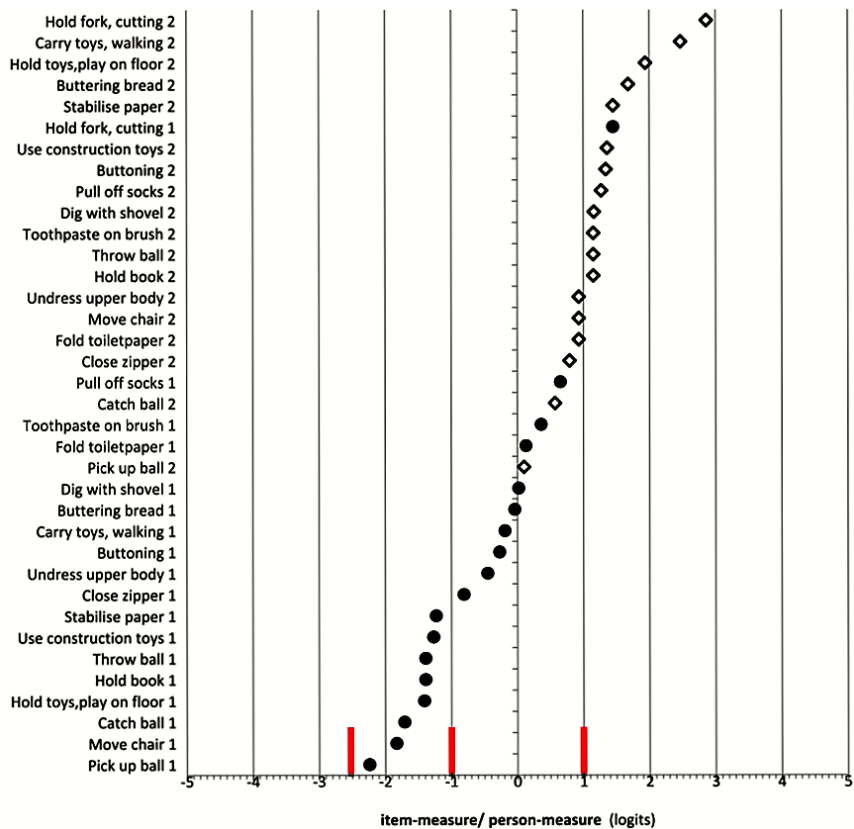
In conclusion, the amount of spontaneous hand-use of a child with unilateral paresis performing daily activities at home can be validly quantified by the HUH. Parents report on their child's spontaneous hand use in 18 bimanual activities using a 5-category rating scale. Item scores are recalculated into a 3-point item-score to obtain a valid sum-score. The HUH seems to be a valuable addition to current clinical assessments. Future studies should (further) establish its construct validity, test-retest reliability, sensitivity to change and should address cross-cultural validation.

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# Appendix

**Appendix 1** Hierarchically ordered item-measures for score 1 (*regularly, often*)(●) and score 2(*always*) (◇) of the 18 HUH items



A child with a HUH-score (person-measure) of +1 logits is likely to obtain a score *regular* or *often* on all items (except 'hold a fork while cutting food') and on some items a score *always* can be expected. A child with a HUH-score of -1 logits has a very low chance to score *always* on any of the HUH items. Children with a HUH-score below -2.5 logits may obtain scores *sometimes* or *never* but are not likely to obtain high item scores for spontaneous use of the affected hand.

## Appendix 2



### Questionnaire

**Questionnaire for parents to measure spontaneous use of the arm and hand in children met unilateral upper limb paresis aged 3 – 10 years.**

Date: ...../...../.....

Child's Name: .....

Date of birth: ...../...../.....

☐ Boy ☐ Girl

Diagnosis:

☐ Unilateral Cerebral Palsy

☐ Obstetric Brachial Plexus Lesion

☐ Other

Affected Side:

☐ Left ☐ Right

**My Child: (Choose the best description for the affected hand)**

☐ picks up small objects like raisins / opens and closes small objects

☐ can pick up certain size objects from the table but not all sizes/ is able to hold an object in the hand

☐ uses arm or hand to support / is unable to grasp / cannot hold objects

**This Questionnaire was completed by:**

☐ Mother ☐ Father ☐ Both parents

☐ Other .....

|  |  |  |  |        |
|--|--|--|--|--------|
|  |  |  |  |        |
|  |  |  |  |        |
|  |  |  |  |        |
|  |  |  |  |        |
|  |  |  |  | logits |



Instructions

In this questionnaire you are asked **how often** your child uses the affected hand while performing the 18 activities.  
The questionnaire consists of bimanual activities but some of them could be performed single handed. The focus of the questionnaire is assess how often your child chooses **to spontaneously use** the affected hand in the activity (so without prompting).

We ask you to choose the answer that is consistent with the regular routine (even if the activity is not often performed). In each question you have 5 or 6 response options.  
**We urge you to mark only 1 option.**

*Your answer options are:*

- Never:** Your child never uses the affected hand performing the activity.  
**Sometimes :** Your child sometimes uses the affected hand performing the activity.  
**Regularly:** Your child uses the affected hand half the time this activity is performed.  
**Often:** Your child often performs this activity using the affected hand.  
**Always:** Your child (almost) always uses the affected hand performing the activity.

Example:

How often does your child use the affected hand spontaneously to assist in **closing a zipper**?  
(jacket/cardigan/pants etc.)

| If you think it is like this:  | Your answer would be: |
|--|-----------------------|
| "My son never closes zippers himself. It is far too difficult for him and that's why we do it for him".  | <b>Never</b>          |
| "My daughter needs help her to close the zipper because that is still too difficult for her. Even though she is able to hold her jacket with one hand and pull the zipper with the other, she seldom does, and if it doesn't work one-handed she almost immediately asks for help" | <b>Sometimes</b>      |
| "My daughter sometimes tries to close the zipper of her jacket but often she does need help. She often uses her affected hand to straighten her jacket when she pulls up the zipper. Still, we help her regularly."  | <b>Regularly</b>      |
| "My son uses his affected hand while opening and closing zippers most of the time. When we are in a hurry or when he is tired we help him".  | <b>Often</b>          |
| "My son independently closes zippers bimanually".  | <b>Always</b>         |

**On the following page you can start answering the questions.**  
**It will take about 10 minutes of your time to fill out this questionnaire.**



### Start Questionnaire

We want to know **how often** your child chooses to **spontaneously** use the affected hand while performing this activity (so without prompting).  
**You can only mark 1 option.**

- 1 How often does your child use the affected hand putting **toothpaste on the toothbrush**?
  - ☐ Never
  - ☐ Sometimes
  - ☐ Regularly
  - ☐ Often
  - ☐ Always
- 2 How often does your child use the affected hand to **hold a toy while playing on the floor**?
  - ☐ Never
  - ☐ Sometimes
  - ☐ Regularly
  - ☐ Often
  - ☐ Always
- 3 How often does your child use the affected hand to assist **undressing the upper body** (remove their singlet, shirt, sweater )
  - ☐ Never
  - ☐ Sometimes
  - ☐ Regularly
  - ☐ Often
  - ☐ Always
- 4 How often does your child use the affected hand to **move a chair**?
  - ☐ Never
  - ☐ Sometimes
  - ☐ Regularly
  - ☐ Often
  - ☐ Always
- 5 How often does your child use the affected hand to **pull off socks**?
  - ☐ Never
  - ☐ Sometimes
  - ☐ Regularly
  - ☐ Often
  - ☐ Always



- 6 How often does your child use the affected hand to **play with construction toys** (lego/duplo/etc.)?
- ☐ Never
  - ☐ Sometimes
  - ☐ Regularly
  - ☐ Often
  - ☐ Always
- 7 How often does your child use the affected hand to assist with **buttering bread**?
- ☐ Never
  - ☐ Sometimes
  - ☐ Regularly
  - ☐ Often
  - ☐ Always
  - ☐ Not applicable due to the age of my child
- 8 How often does your child **hold a toy** in the affected hand while **walking**?
- ☐ Never
  - ☐ Sometimes
  - ☐ Regularly
  - ☐ Often
  - ☐ Always
- 9 How often does your child use the affected hand to **hold a booklet** while **reading or looking at pictures**?
- ☐ Never
  - ☐ Sometimes
  - ☐ Regularly
  - ☐ Often
  - ☐ Always
- 10 How often does your child use the affected hand to assist with **tearing off and folding toilet paper** when using the toilet?
- ☐ Never
  - ☐ Sometimes
  - ☐ Regularly
  - ☐ Often
  - ☐ Always



Please keep in mind that we want to know how often your child chooses to **spontaneously use** the affected hand while performing this activity.  
**You can only mark 1 option.**

- 11 How often does your child **throw a large ball** (football size) with both hands while playing ball games?
  - ☐ Never
  - ☐ Sometimes
  - ☐ Regularly
  - ☐ Often
  - ☐ Always
- 12 How often does your child use the affected hand to **pick up a large ball** (football size) from the floor?
  - ☐ Never
  - ☐ Sometimes
  - ☐ Regularly
  - ☐ Often
  - ☐ Always
- 13 How often does your child use the affected hand to **hold a fork while cutting food**?
  - ☐ Never
  - ☐ Sometimes
  - ☐ Regularly
  - ☐ Often
  - ☐ Always
  - ☐ Not applicable due to the age of my child
- 14 How often does your child use the affected hand to **stabilise paper while drawing or writing**?
  - ☐ Never
  - ☐ Sometimes
  - ☐ Regularly
  - ☐ Often
  - ☐ Always
- 15 How often does your child use both hands together to **catch a large ball** (football size) while playing ball games?
  - ☐ Never
  - ☐ Sometimes
  - ☐ Regularly
  - ☐ Often
  - ☐ Always





- 16 How often does your child use the affected hand to **hold a shovel** while **digging a hole in the sand** (beach/garden/sandbox)?
- ☐ Never
  - ☐ Sometimes
  - ☐ Regularly
  - ☐ Often
  - ☐ Always
- 17 How often does your child use the affected hand to assist with **opening and closing buttons** on their clothes? (jacket/blouse/pants etc.)
- ☐ Never
  - ☐ Sometimes
  - ☐ Regularly
  - ☐ Often
  - ☐ Always
- 18 How often does your child use the affected hand to assist in **closing a zipper**? (jacket/cardigan/pants etc.)
- ☐ Never
  - ☐ Sometimes
  - ☐ Regularly
  - ☐ Often
  - ☐ Always

Thank you, this was the last question.





# 8

## Summary and General Discussion



## Summary

### General Introduction

With a prevalence of 2 to 2.5 per 1000 living births cerebral palsy (CP) is one of the most common causes of motor impairments in childhood. About 30 % of this population is classified with unilateral spastic CP, indicating that they have motor impairments mainly on the right or left side of the body. In most cases the upper limb is more involved than the lower limb. The effective use of the arm and hand to reach for, grasp, hold, release and manipulate objects with the affected upper limb is often compromised and hampers the execution of bimanual activities in daily life. However, even when upper limb motor control is fairly good, there are children with unilateral upper limb paresis that show a marked discrepancy between their upper limb capacity<sup>1</sup> (i.e., what they can do with their affected hand in optimal conditions) and their performance<sup>1</sup> (i.e., how they actually use the affected hand performing daily activities). This 'underuse' of the affected upper limb is referred to as 'developmental disregard' (DD).

Based on findings in experimental research with monkeys and on studies with adult stroke patients, new treatments protocols have been developed that aim to improve upper limb motor performance in children with unilateral CP. Both constraint induced movement therapy (CIMT) and bimanual training (BiT) are protocols consisting of short intensive periods of massed upper limb training. The aim is to improve upper limb capacities, optimize bimanual coordination, and decrease developmental disregard with the ultimate goal to improve the performance of daily activities. The most striking difference between these interventions is that during CIMT the non-affected upper limb is restrained by a sling, cast or splint while the affected arm and hand are intensively trained, whereas in BiT the affected limb is intensively trained during bimanual activities without any restraint. In order to improve its feasibility and efficacy in young children (2.5 to 8 years) we modified the original CIMT protocol (mCIMT) and combined it with BiT to enhance the transfer of improved upper limb movements into age-appropriate bimanual activities. We also adjusted this mCIMT-BiT protocol for older children with unilateral CP (aged 8 to 18 years) by incorporating self-management training within the BiT element, in order to reinforce the children to independently monitor the efficient use of their affected hand during daily life activities without being prompted by adults. The combined use of mCIMT and BiT within one protocol was a novel approach and the effectiveness of this combined therapy approach was not tested before. By the same token, the combination of mCIMT-BiT with self-management training was novel and had not been tested yet.

<sup>1</sup> Terminology according to the international classification of functioning, disability and health in children and youth (ICF-CY)

Despite the fact that effects of intensive upper-limb interventions are assessed with valid and reliable outcome measures, some aspects of capacity and performance cannot be assessed specifically enough, because the existing measures lack detail. More specifically, in young children (2.5-8 years) the capacity to grasp, hold and release objects cannot be assessed adequately with the current capacity tests as too few items address the manipulation of objects. Alternative tests that do measure the capacity to grasp, hold and release require the child to perform 'as quickly as possible' and are, thus, not very suitable for this age group. Hence, we examined whether the recently published Modified House Classification (MHC), which assesses the capacity to grasp, hold and release objects through 32 items without time constraints, could be used in children with unilateral CP. Regarding the assessment of upper limb performance, we specifically identified a lack of instruments that can validly and reliably assess the amount of spontaneous hand use in children with unilateral CP. Therefore, we refined and revised the Video Observation Aarts and Aarts-Determine Developmental Disregard (VOAA-DDD). The VOAA-DDD measures differences in capacity, performance and amount of hand use (to determine developmental disregard) in two bimanual tasks. Validity and test-retest reliability of the revised VOAA-DDD were assessed. An inherent shortcoming of clinical tests is that they are not designed to measure spontaneous hand use in natural and daily-life circumstances. To fill this important gap, we developed a new parent-rated questionnaire and tested its clinimetric properties.

This thesis starts with a general introduction (Chapter 1) providing background information on children with unilateral CP, intensive upper-limb therapy, and on the assessment of upper-limb capacity, performance and amount of spontaneous hand-use. The remainder of this thesis consists of two main parts. Part one focusses on upper limb interventions in unilateral CP. First, it presents the studies examining the effectiveness of the mCIMT-BiT intervention in young children. Second, a feasibility study of the CIMT-BiT intervention augmented with self-management training in older children with unilateral CP is presented. Part two addresses upper limb assessment in unilateral CP. In this part, the revisions of two observational scales (the MHC and the VOAA-DDD, respectively) are described and the clinimetric properties of their revised versions are examined. The third study presents the development of the Hand-Use-at-Home Questionnaire (HUH), as well as the examination of its internal structure, unidimensionality and validity.

## **Part one: Upper limb interventions in unilateral CP**

### **Chapter 2**

In this chapter we report an effectiveness study of a new playful upper limb intervention for young children with unilateral spastic CP. This mCIMT-BiT intervention (i.e., the Pirate group intervention) started with 6 weeks modified Constraint Induced Movement Therapy (mCIMT) followed by 2 weeks of Bimanual Training (BiT). A randomized controlled

trial (RCT) was used to investigate whether the mCIMT-BiT intervention improves the spontaneous use of the affected limb, both qualitatively and quantitatively compared to usual care (UC) of the same duration. Children with unilateral spastic CP, aged 2.5 to 8 years and with Manual Ability Classification System (MACS) scores I, II, or III, were recruited and randomly allocated to either the mCIMT-BiT group (i.e. the intervention group) or the UC group (control group).

The mCIMT-BiT (n=28) group weekly received three days of 3-hour trainings sessions (9 hours each week). In the first six weeks they received 54 hours of mCIMT, followed by two weeks of task-specific training in goal-directed bimanual play and self-care activities (18 hours). Usual care (n=24) consisted of 1.5 hours of general physical or occupational therapy weekly plus daily training (with a minimum of 7.5 hours a week) focused at optimal use of the affected hand by the parents at home and the teacher at school (9 hours each week). Primary outcome measures were the Assisting Hand Assessment (AHA) and the ABILHAND-Kids. Secondary outcome measures were the Melbourne Assessment of Unilateral Upper Limb Function (Melbourne), the Canadian Occupational Performance Measure (COPM), and the Goal Attainment Scale (GAS). Except for the Melbourne, all primary and secondary outcome measures demonstrated significant improvements in the mCIMT-BiT group which were retained at 8 weeks follow-up. In contrast, the control group did not show significant improvements over time. We concluded that mCIMT followed by task-specific training of goal-directed bimanual play and self-care activities is an effective intervention to improve the spontaneous use of the more affected upper limb in children with unilateral CP with relatively good upper extremity function at baseline.

### Chapter 3

To determine long-term effectiveness of the interventions all children that participated in the RCT described in Chapter 2 were re-assessed at 6 months post intervention. The primary (AHA, ABILHAND-Kids) and secondary outcome measures (Melbourne, COPM) were compared between groups. Additionally, children of the intervention group were assessed again 1 year after the intervention, although 5 of them were lost in the follow-up. We found that performance was stable on all primary outcome measures in both groups at six months, while the results on the primary outcomes in the mCIMT-BiT group (n=23) were stable even one year following the intervention. Retention of effects after six months post intervention was also found for the satisfaction and performance scores of the five most important daily life problems as assessed with the COPM. Unilateral capacity as measured with the Melbourne showed a significant improvement in the mCIMT-BiT group between one week and six months post intervention. Our findings were coherent with other studies that compared the results of CIMT with BiT. Our study demonstrated that 72 hours of mCIMT-BiT leads to similar retention of results compared to CIMT or BiT interventions of 90 hours.



The second aim of this study was to register the progression of manual dexterity during 6 weeks of mCIMT (54 hours) followed by 2 weeks BiT (18 hours) and to establish the maximal training effect during the intervention. In addition, potential factors influencing motor learning were assessed. The goal was to extract information based on which the optimal dosage of intensive upper-limb training could be determined. All children allocated to the mCIMT-BiT group ( $n=28$ ) were assessed weekly on the Box and Block test (BBT). The number of blocks transferred with the affected hand were counted and the raw scores were used to compose individual learning curves. Maximum training effect was independently determined by 2 blinded raters and was defined as the moment that the top of the learning curve was reached. Fifteen children (53.6%) reached a maximum training effect within the mCIMT period (6 weeks). This group of children differed from the others with respect to age, but not gender, affected side, or manual ability. Children younger than five years had a greater chance to reach a maximum score (BBT) within 6 weeks mCIMT. This maximum stabilized already after four weeks. Older children showed a longer progression and tended to decline afterwards during the BiT part of the program. These findings suggest that children of 5 years and older might benefit from a longer period of mCIMT than 54 hours to reach their maximum unimanual capacity and to retain this capacity during subsequent bimanual training.

## Chapter 4

High intensity upper-limb therapy for older children and adolescents with unilateral CP (8-18 years) are generally restricted to day-camp models during school holidays. Until now, day-camps with only one type of intervention (CIMT or BiT) have been described for this group. These camps provide 30 to 90 hours of training during one to three weeks. The exact critical dose of CIMT or BiT to achieve meaningful changes in either unimanual capacity or bimanual performance is still a matter of debate. Adding to this debate we examined whether a combined CIMT-BiT intervention of one week would be feasible and effective to improve both unilateral upper limb capacity and the performance of bimanual activities in older (8-18 years) children with unilateral CP. The intervention would be guided by individual goal-setting. Self-management training was added to the CIMT-BiT intervention to maximize the effect of training and to empower the participants with respect to self-monitoring the effective use of their affected hand.

Functional goals (COPM), unimanual capacity (Box and Block test), bimanual performance (ABILHAND-Kids, Children's Hand-use Experience Questionnaire (CHEQ) and amount of use (VOAA-DDD-R) were measured at baseline, one week and four months post intervention. Twenty children (mean age 9.5 years) participated in the intervention. Compared to baseline, there were significant improvements on all outcome measures. The largest effect sizes were found for the COPM-performance and COPM-satisfaction (Cohen's  $d=2.09$  and  $d=2.42$ , respectively). The effect size was large for the ABILHAND-Kids ( $d=0.86$ ), moderate for the CHEQ ( $d=0.70$ ) and Box and Block test ( $d=0.56$ ), and small for

the VOAA-DDD-R ( $d=0.33$ ). All effects were retained at the four months post intervention assessment. The results of this study indicate that one week (36 hours) of intensive mCIMT-BiT combined with self-management training is a feasible and promising intervention for improving the capacity of the upper limb and its use in bimanual activities in older children and adolescents with unilateral CP.

## Part two: Upper limb assessment in unilateral CP

### Chapter 5

Children with unilateral CP have a reduced capacity to handle objects with the affected hand and, consequently, are hindered in the performance of bimanual activities. Several instruments are available for the assessment of upper limb capacity in children with unilateral CP. However, a validated instrument that specifically evaluates the maximal capacity of the affected hand to grasp, hold and release in bimanual tasks does not exist. The MHC was introduced by Koman et al.<sup>2</sup> with 32 items related to functional upper limb use. The sum score (i.e., number of passed items) represents the functional capacity of the assessed hand (range 0-32 points). In our study we examined whether the MHC can be used to objectively discriminate between levels of functional capacity of the affected hand in children with unilateral CP. Because the MHC was not specifically developed for children with unilateral CP, we evaluated its unidimensionality and item hierarchy in a large cohort of children with unilateral CP (calibration cohort).

MHC items were scored from 369 videotaped assessments of 159 children with a median age 6.5 years (range 2.1 – 17.4 years). Rasch analysis was used to improve clinimetric properties by eliminating, re-ordering and weighting items. We found that fifteen of the 32 MHC items could be included in the Rasch analysis. The excluded items were either too easy or too difficult for our sample. Fourteen items worked well together to measure functional capacity. The Rasch-reduced 14-item set was then tested for its construct validity within the calibration cohort as well as in another sample of 40 children (median age 8.1 years) with unilateral CP (validation cohort). We compared the weighted sum score to the MACS and to the ABILHAND-Kids. In the validation cohort the 14-item MHC had a strong correlation with the MACS and a fair correlation with ABILHAND-Kids.

We concluded that the original item hierarchy of the MHC can be improved in order to use its sum score for the assessment of upper limb capacity in children with unilateral CP. The Rasch-reduced 14-item MHC with weighted sum score shows good construct validity to measure functional capacity of the affected hand in children with unilateral CP.

2 Koman LA, Williams RM, Evans PJ, Richardson R, Naughton MJ, Passmore L, et al. Quantification of upper extremity function and range of motion in children with cerebral palsy. *Dev Med Child Neurol.* 2008; 50: 910-7

## Chapter 6

In this study the validity and reliability of the VOAA-DDD-R was evaluated. This test aims to assess capacity, performance, and developmental disregard of the affected upper limb in children with unilateral CP. It consists of two standardized bimanual tasks that are recorded on video and are scored off-line. One task demands the use of both hands (stringing beads), while the other merely stimulates the use of both hands (decorating a muffin). In this revised version of the VOAA-DDD the scoring system was simplified and the tasks were adapted to improve the contrast between both tasks (i.e., in eliciting hand use) and to improve the similarities in observable activities of the affected arm and hand. The frequency of grasping, holding and releasing was assessed irrespective of the quality of movement as well as the duration of use of the affected upper limb during each task. Three scores were derived from the assessment. The frequency score of the 'demanding' beads task was used to calculate the capacity score (0-100%) and the frequency score of the 'stimulating' muffin task was used to calculate the performance score (0-100%). The difference in duration of use of the affected upper limb between both tasks was used as a measure of developmental disregard (0-100%). Twenty-five children with unilateral CP aged 2.5 – 8 years and 46 age- and gender-matched typically developing children participated in the study. The VOAA-DDD-R was administered twice. Construct validity was determined by comparing children with CP with typically developing children. Intra-rater, inter-rater, and test-retest reliability were determined using the intraclass correlation coefficient (ICC). Standard error of measurement (SEM) and smallest detectable differences (SDD) were calculated additionally. The cut-off score that was used to determine developmental disregard was set at the mean score +2 SD of typically developing children. We found that children with unilateral CP scored significantly lower compared to typically developing children on capacity (77% vs 98%, respectively) and performance (55% vs 100%, respectively), yielding much higher scores on developmental disregard (23% vs 7%, respectively). The ICCs were high (0.79–1.00) and indicated good reliability. Standard errors of measurement ranged from 4.5 to 6.8% and smallest detectable differences ranged from 12.5 to 19.0%. We concluded that the three scores of the VOAA-DDD-R (i.e., capacity, performance, and developmental disregard) can reliably and validly be used to assess the affected upper limb in children with unilateral CP. Results confirm that many children with unilateral CP show a discrepancy between what they 'can do' with their affected upper limb when bimanual activity is demanded (i.e. capacity) and what they 'actually do' when bimanual activity is merely stimulated (i.e. performance). The VOAA-DDD-R scores can thus be used to uncover specific problems in upper-limb functioning of children with unilateral CP and give direction to individually tailored upper limb therapy.

## Chapter 7

In both unilateral CP and Neonatal Brachial Plexus Palsy (NBPP), repeated failure to perform activities with the affected arm and hand may lead to a decreased spontaneous use of this hand in daily activities. As a consequence, children with a unilateral paresis often use their affected upper limb less frequently in bimanual activities than might be expected based on their functional capacities. Despite intensive rehabilitation, parents often report that their children still underuse the affected arm and hand at home and perform many activities unimanually. To date, no valid and reliable measure exists that assesses how often children with unilateral upper limb paresis actually use their affected arm and hand in daily life. Still, underuse of this limb is recognized as an important problem. Therefore, a parent-reported measure to assess the amount of spontaneous hand use during every day bimanual activities provides a valuable addition to the existing clinical assessments.

In this chapter the development of the Hand-Use-at-Home questionnaire (HUH) is described. In addition, its internal structure and construct validity are examined. The HUH evaluates how often the affected upper limb is used in typical daily life activities of children with unilateral upper limb paresis, aged three to ten years. Parents rated the spontaneous use of the affected hand of their child on a 5-point rating scale for each activity. The sum-score of the HUH was expected to reflect the extent to which the child spontaneously used the affected hand at home. To be able to validly sum the item-scores, we tested the rating-scale structure, the unidimensionality of the item set and the hierarchy of the items with Rasch analysis. To examine internal validity, data of 322 children (mean age 6.6 years, SD 2.1 years) with unilateral CP (n=131) or NBPP (n=191) were collected. Additionally, data of 55 typically developing children (TD) (6.8 years, SD 2.5 years) were used to examine construct validity. Rating scale analysis showed that the 5-point rating scale was disordered in all items. Therefore, it was collapsed into a well-structured 3-point rating scale. The results showed that 18 hierarchically ordered bimanual activities, scored on a 5-point rating scale and recalculated into a 3-point rating scale, fitted the Rasch model within acceptable range. Each item (activity) had two item-measures: the lowest for score 1 (i.e., the degree to which the activity elicited a score *regular* or *often* for spontaneous use of the affected hand) and the highest for score 2 (indicating the degree to which the activity *always* elicited use of the affected hand). The spread of the item-measures indicated that the activities were adequately eliciting spontaneous hand use ranging from 'easy' to 'difficult'. The HUH-scores ranged from -4.69 to 5.17 logits. Internal consistency of the HUH was high (Cronbach's  $\alpha = 0.941$ ).

The HUH sum score was able to discriminate between children with unilateral CP, children with NBPP and typically developing children supporting the construct validity of the HUH. The TD group had the highest amount of spontaneous hand use (median: 2.51 logits) with 5.5% of the children obtaining the maximum score. The lowest median HUH-score was for the unilateral CP group (-0.343 logits). The NBPP group had an intermediate median HUH-score of 1.07 logits.

It was concluded that the HUH is a valid measure to assess the amount of spontaneous affected hand use in children with unilateral upper limb paresis, aged three to ten years old. Future studies establishing test-retest reliability and sensitivity to change of the HUH are warranted.

## General discussion

In this thesis the effectiveness of a novel upper-limb intervention was assessed for children with unilateral cerebral palsy (CP) by *combining* (modified) constraint induced movement therapy (CIMT) with bilateral training (BiT). The intervention was specifically tailored to the needs of a younger (2.5 to 8 years) and an older (8 to 18 years) age group. In the latter group, self-management techniques were added to achieve optimal results in a limited period of training. Because the age of a child is an important determinant of the content of interventions in pediatric rehabilitation as well as of the choice of outcome measures, several aspects and findings of the studies in this thesis will be discussed by taking into account the influence of age.

### CIMT-BiT ratio

The combination of CIMT and BiT to optimize upper limb motor behavior in children with unilateral CP attempts to enhance the integration of newly acquired upper limb skills with relevant bimanual activities and individual goals. During the last decade, many studies have examined the effectiveness of CIMT or BiT, from which combined approaches have emerged. Dose-matched comparisons of CIMT versus BiT demonstrated similar gains in upper limb outcomes.<sup>1</sup> However, a recent systematic review<sup>2</sup> showed that there is still a large variation in the dosage and intensity of training and indicated that a dosage of at least 40 hours of therapy is needed to reach meaningful clinical changes. Still, the optimal dose of training is a matter of debate<sup>1,2</sup>, but will most likely be between 40 and 60 hours of training.<sup>3</sup> Cumulating evidence suggests that both CIMT and BiT are superior to usual care to improve the movement capacity of the affected upper limb as well as bimanual hand use.<sup>1</sup> However, where CIMT seems to promote unilateral upper limb capacity more than BiT, BiT seems to be favorable for promoting bimanual coordination and goal attainment. In addition, some concerns<sup>4</sup> exist with regard to the functional development of the non-affected side in (young) children when CIMT is applied in isolation.

In this thesis we assessed the effectiveness of two specific upper limb interventions for children with unilateral CP combining CIMT with BiT (chapters 2 and 4). Both interventions focused on improving upper limb capacity, bimanual performance and the amount of use of the affected arm and hand. The interventions were specifically tailored to the needs of both age groups (2.5 to 8 years and 8 to 18 years) and proved to be effective. However, the total dosage and intensity of training as well as the CIMT-BiT ratio varied substantially. In the Pirate-group intervention (chapter 2) consisting of 72 trainings hours (9 hours per week), the CIMT-BiT ratio was 75-25% with the main focus on improving upper limb capacity and reduction of developmental disregard during the CIMT period. This intervention yielded good results, superior to usual care, and was feasible even in very young children. Interestingly, the findings regarding the effect of age on motor learning (chapter 3) provided some indication that the applied CIMT-BiT ratio was not yet optimal

for the children of 5 years and older. Dexterity in the children aged between 2.5 and 5 years improved from the start of the CIMIT intervention and already stabilized after 4 weeks (36 hours), whereas the scores of the children aged 5 to 8 did not plateau in the CIMIT period and decreased somewhat during the 2 weeks (18 hours) of bimanual training. Perhaps older children (5-8 years) might benefit from a longer period or higher intensity of unilateral training to reach and stabilize their maximal upper limb capacity. The intervention for the older children (chapter 4) consisted of 40 hours of therapy in 5 days and used a CIMIT-BiT ratio of 37.5-62.5%. Based on a pilot study (unpublished) we decided that the amount of BiT should exceed the amount of CIMIT in order to provide sufficient opportunities to train bimanual activities and in order to implement concurrent self-management training. Each day started with 3 hours of CIMIT to 'refresh' or specifically train necessary skills, which was followed by 5 hours of bimanual training. It is, nevertheless, unclear how our results relate to the provided CIMIT-BiT ratio and to what extent the self-management training influenced the effects. Only a few studies examining CIMIT-BiT interventions have been published.<sup>5-7</sup> Two small studies with young children favored CIMIT over BiT and applied 24 hours per day restraint of the unaffected upper limb in addition to training of the affected upper limb using CIMIT-BiT ratios of 93-7%<sup>5</sup> and 86-14%<sup>6</sup>. A study providing a 45-hour (2 weeks) hybrid CIMIT intervention with a 50-50% CIMIT-BiT ratio for children between 5 and 16 years<sup>7</sup> yielded sustained improvements on unilateral capacity (dexterity) and scores for individual goals, but the improvements on bimanual performance were not clinically meaningful in most children and were not retained. The authors argued that their therapy dose had probably been insufficient to achieve clinically meaningful results on bimanual performance. Our CIMIT-BiT intervention for older children, however, showed that with 36 hours of intensive CIMIT-BiT training sustained results can be achieved. This discrepancy may be due to the fact that we applied a different CIMIT-BiT ratio (37.5-62.5%), a 1:1 therapist to child ratio, and included self-management training. Until now, no studies have been conducted that compared CIMIT or BiT to a combined CIMIT-BiT intervention. Consequently, as of yet, no definite conclusions can be drawn on the efficacy of CIMIT-BiT intervention compared to CIMIT or BiT alone, nor on the optimal CIMIT-BiT ratio. Given the state of evidence thus far (i.e. CIMIT and BiT yield comparable results<sup>8-10</sup>), the most favorable approach is to choose an intervention that fits most closely to the individual characteristics and age of the child and the goals and preferences of the child and its parents.

### Goal setting

Individual goal setting is an important component of task-specific training<sup>11</sup> because it enhances performance and motivation.<sup>12</sup> This has been emphasized in studies on the effects of mastery motivation (i.e., the intrinsic force that directs our persistence and intention to pursue challenging tasks or activities) on goal attainment.<sup>13-16</sup> Hence, collaborative goal setting with families is a key component of intensive upper limb

interventions.<sup>1</sup> Several studies have shown that caregivers may have a different perspective on their child's functioning than the children themselves and that parents' goals for intervention often differ from those of their children.<sup>17, 18</sup> In the one-week intervention for older children, in which we taught them to self-manage the use of their affected hand, self-determined goals were considered an important element. In contrast to previous interventions where parents determined goals on behalf of their (young) child, in this intervention the children and adolescents were actively involved in goal setting using the COPM. For many parent-child dyads it was a novel experience that preferences of the children prevailed and parents were often pleasantly surprised by their child's choices and reasoning. The children prioritized up to ten goals and most of them ranked the 3 most important goals independently or with the help of the assessor or the parents available as 'consultants'. Selected activities often related to the aspiration to participate in the social environment with peers. More than 60% of all goals was related to self-care, 16% to productivity (school, household chores) and 21% to leisure. Most children motivated their choices as follows: "I know this goal is important for my mom, but for me throw up the ball to serve with tennis is more important", "I want to cut my meat independently so I don't need help when I eat at my friend's house" and "I'd like to give a 10 for performance when washing my hair independently but the truth is that my mom still needs to check if all the soap is rinsed out, so it is still a 7". All participants were well aware of their goals during the intervention and showed motivation to practice, although we did not specifically examine mastery motivation as was suggested in the literature.<sup>14, 16</sup> Our participants not only achieved meaningful changes on the 3 most important goals, which was reflected in the large effect sizes for COPM-Performance and COPM-Satisfaction (2.09 and 2.42, respectively), but achieved satisfactory results on almost all other goals as well, indicating that all selected goals were achievable.<sup>19</sup> The large effect sizes and retained results on the ABILHAND-Kids and COPM reflect the children's increased level of independency that could only be reached by engagement in therapy and high levels of persistence to practice during and after the intervention. The COPM is not applicable for children younger than 8 years.<sup>20</sup> Therefore, in the Pirate-group intervention, we used the COPM with the parents to set achievable goals that represented age-appropriate activities, important for the child and/or for its parents. Based on the notion that priorities and perspectives can differ between parents and children,<sup>17, 18</sup> the Perceived Efficacy and Goal Setting System (PEGS) was developed for children with disabilities aged 6 to 9 years.<sup>17, 20</sup> The PEGS uses visual cues to narrow goal areas, to enable children to report on their competence to perform daily activities and to identify goals. The self-identified goals were found to be achievable to the same extent as goals identified by parents,<sup>19</sup> although children prioritized different goals for the intervention. Given these results, it is advisable to involve children aged 5 years and older in the process of goal setting in future CIMT-BiT interventions.



### **Self-management training**

The use of self-management techniques was a novel addition to intensive upper-limb training. We applied self-management training to maximize the intervention effects. Additionally, we aimed to empower the children to take responsibility for self-monitoring the (supportive) use of their affected upper limb during daily activities and to become less dependent on being prompted by adults.

Self-management usually refers to a much broader perspective of someone's functioning and relates to the tasks that an individual must undertake to live well with one or more chronic conditions<sup>21</sup>. Self-management interventions start from the clients' perspective and support individuals to develop knowledge, skills and confidence to manage some or all aspects of medical, emotional and/or social role.<sup>22</sup> In our intervention for older children we implemented a single aspect of the broader concept of self-management aiming at a specific part of the child's functioning: to independently focus on effective use of the affected upper limb in bimanual activities without being prompted by others. This aspect was chosen from the adult perspective rather than from the child's perspective. A behavioral approach was used to teach the children to complete self-management steps (e.g., self-instruction, self-monitoring, self-evaluation and rewarding). The intervention provided parents and children with tools for activity planning and problem solving and increased the awareness of the different ways the children could use their affected upper limb for independent task execution. Still, the aspect 'problem solving' will possibly need more attention in future interventions, as many older children spontaneously applied for the 1-week intervention two or three years in a row to work on new goals. Supporting self-management of the consequences a chronic condition such as unilateral CP is an ongoing process,<sup>22</sup> especially for adolescents in transition to adulthood. The question emerges how we can prepare adolescents with unilateral CP, and their parents, to deal with future challenges regarding upper limb use without the immediate need to rely on the knowledge of a therapist. Knowledge, skills and confidence to solve problems cannot be acquired through education alone,<sup>23</sup> but can be gained through additional self-management training directed at problem-solving skills. These include problem definition, generation of possible solutions, use of social and professional resources, solution implementation, and evaluation of results.<sup>24-26</sup>

Throughout childhood, pediatric upper limb interventions for a child with CP may need to shift from optimizing capacities and performance to optimizing 'self-solving' abilities. A group intervention for adolescents addressing problem-solving skills in a broader perspective than the functional use of the affected upper limb alone is likely an important next step.

### **What improves after CIMT-BIT?**

In our upper limb interventions we aimed to increase upper limb capacities and daily use and to promote integration of newly acquired upper limb skills into bimanual activities

and individual goals. The ultimate goal was to decrease or even overcome developmental disregard and thereby achieve automatic, spontaneous use of the affected hand as an assist in daily activities. Collectively, the results of the interventions described in chapters 2 and 4 showed that the participating children improved their upper limb capacities and affected hand use in bimanual activities. They also achieved more functional independence concerning specific, individual goals. A secondary analysis of the Pirate-group intervention<sup>27</sup> examined whether the improvements on activity level could be attributed to better active and/or passive range of joint motion on the level of body functions and structures of the International Classification of Functioning, Disability and Health (ICF).<sup>28</sup> Post-intervention results showed that the mCIMT-BiT intervention did not lead to significant gains in passive or active elbow or wrist extension, although there was a positive trend towards improvement (but not retained). This implies that the observed improvements at activity level cannot be explained by underlying improvements of joint or neuromuscular functions, suggesting a more effective utilization of *existing* motor functions of the affected arm and hand rather than true neuromuscular repair. Children with *moderate* wrist control at baseline showed the largest improvements on capacity scores of the VOAA-DDD, while children with a MACS II and III classification at baseline improved most on performance scores of the VOAA-DDD. These results are in line with other studies where larger gains after CIMT were associated with greater impairments at baseline<sup>1</sup>. Interestingly, children with a MACS III classification showed the largest decrease in DD-scores of the VOAA-DDD (reflecting the discrepancy between capacity and performance), although this effect was not retained. Likewise, with regard to the 1-week intervention for older children, four out of eight (50%) of the participants with DD at baseline still had DD post intervention (chapter 4). Thus, our hypothesis that the CIMT-BiT interventions would result in a significant and long-lasting reduction of developmental disregard in children with unilateral CP was not corroborated.<sup>27</sup>

A possible explanation for the limited effect of our CIMT-BiT interventions on developmental disregard as assessed with the VOAA-DDD-(R) is that, in spite of the improved upper limb capacities and performance, insufficient automaticity was reached to obtain an optimal degree of spontaneous arm-hand use during bimanual activities.<sup>27</sup> Similar to learning skills like playing the piano in typically developing children, automated hand use during bimanual tasks in children with unilateral CP may take years of training and practice. Hung et al.<sup>29</sup> showed that, even when children with unilateral CP learn to perform a simple bimanual task, they improve more slowly compared to typically developing children and they need 2 to 3 times more practice. Hence, fully automatic use of the affected hand in daily bimanual activities may require a much longer training period than a upper limb intervention can offer. From a neuropsychological perspective, Houwink et al.<sup>30</sup> proposed that the attentional demands of affected hand use in bimanual tasks, especially during the first 'cognitive' phase of learning as described by Fitts and

Posner,<sup>31</sup> may be an explanatory factor for underuse of the affected upper limb in children with unilateral CP. Evidence for increased attentional demands was found in children with developmental disregard specifically when preparing a response with the affected hand in a (bimanual) dual task situation.<sup>32</sup> This finding suggests that to enable automatic use of the affected hand in bimanual activities, upper limb training needs to aim for a decreasing need of attentional resources as well. Thus, training may start in a therapeutic setting aimed at problem-solving and promoting self-initiated upper limb use in relatively simple situations, but gradually needs to shift towards bimanual performance in a more complex and distracting environment. Future studies are warranted to find evidence whether such an approach will better attain and consolidate any improvements in developmental disregard. Adequate automaticity of affected hand use in daily life will also require practice and stimulation in the home environment. This, however, will put an extra burden on the time and energy of parents who are already heavily involved in care giving, enabling participation<sup>33</sup> and coordinating services. Increasing parental stress may lead to lower compliance to home-based programs.<sup>34, 35</sup> Based on the expectation that automatic use of the affected hand needs a long period of training and stimulation with meaningful and progressively challenging activities, a static advice of activities to practice will not suffice; a dynamic way of coaching parents and children may be needed to optimize upper limb use in daily life. Therefore, parents and children need to be an integral part of the treatment team such that individualized services and tools for age-appropriate practices at home can be offered. Telephone or video consultation, apps with stimulating activities or specific challenges for children, but also home-based programs mimicking the CIMT-BiT intervention and providing booster-sessions for children to enhance affected hand use and practice new goals, may be helpful to finally overcome developmental disregard.

## Assessment

A problem inherent in any intervention study is whether outcome assessment is based on the appropriate outcome measure(s). In the introduction it was already stated that, to evaluate the effect of a pediatric upper limb intervention, an assessment needs to consist of a balanced set of reliable tests that have been validated for the study population taking into account age and diagnosis. Many instruments have been published that could be used to measure specific aspects of upper limb capacity and/or performance in children with unilateral CP, however, not all available instruments are proven reliable and valid for this group (e.g. Jebson Taylor test,<sup>36</sup> Peabody Developmental Motor Scales (2<sup>nd</sup> edition) (PDMS-2),<sup>37</sup> Bruininks–Oseretsky Test of Motor Proficiency (2<sup>nd</sup> edition)<sup>38</sup>). Using such instruments as an outcome measure in upper limb intervention studies does not provide meaningful information on the effectiveness of these interventions, unless sound psychometric properties for the target group are (later) established in studies with adequate sample sizes. For example, the PDMS-2 is a norm-referenced test for typically developing children. It consists of many bimanual items as a result of which children with

unilateral CP are unable to meet the criteria. The same applies to instruments that are not validated for a child's age. Systematic reviews evaluating upper limb measures and their psychometric properties can be very helpful to select the best outcome measures for an intervention study.<sup>39-42</sup> Fortunately, nowadays, more and more test-developers update or revise their instruments and publish additional psychometric data<sup>43,44</sup> so that these can be included in systematic reviews. A basic principle should be that new instruments must be developed only when the available instruments fail to measure the essential aspects that are addressed by an (upper limb) intervention. Indeed, the downside of adding new (disease-specific) outcome measures is that comparisons between and pooling of data across upper limb intervention studies will become increasingly difficult. In 2013 a consensus-group reviewing the evidence on CIMT and BiT counted 48 different outcome measures used to evaluate these interventions.<sup>45</sup> Against this background, the development of a core set of instruments with sound psychometric properties to assess the most important aspects of upper limb functions, capacities and performance could be helpful to guide the conduct of future intervention studies for the sake of homogeneity and comparability.

Probably the most important feature of an outcome measure is the construct it pertains to and how well this construct matches the intervention goals. In addition, an outcome measure should be sufficiently responsive to the intervention. For instance, to evaluate improvement of upper limb capacities after botulinum toxin injections in the elbow or shoulder region a manual dexterity test (e.g. the Box & Blocks test<sup>46</sup>) will probably not be suitable, whereas the Melbourne<sup>47</sup> (measuring the quality of movements based on many 'shoulder-items') may be much better. With regard to our CIMT-BiT interventions (with the main focus on improving object-related hand use), the Melbourne did not detect immediate post-intervention improvement, whereas the 14-item MHC (assessing functional capacity of the hand) was able to detect such improvement. In chapter 4, the 14-item MHC was used as secondary outcome for upper limb capacity and we found a small but significant change compared to baseline. In addition, in 20 young children (2.5-7 years, median age 4.3 years) participating in a recent Pirate-group intervention (n=2 MACS I, n=9 MACS II, n=9 MACS III) we found significantly higher immediate post-intervention scores for the 14-item MHC (median difference 0.29 logits, range 0 – 0.73 logits,  $p<0.001$ ) compared to baseline (unpublished data). This is a first proof that the 14-item MHC, in contrast to the Melbourne, is actually able to measure immediate post-intervention improvement of upper limb capacity after an intensive upper limb intervention. It suggests that an instrument specifically aimed at assessing object-related hand skills (e.g. grasping, releasing) may be more suitable to measure differences after a CIMT-BiT intervention than a more generic upper limb test, such as the Melbourne or the Quality of Upper Extremity Skills Test,<sup>48</sup> particularly for younger children. In older children, dexterity tests measuring speed and accuracy (e.g. Jebson Taylor) seem to effectively measure changes in upper-limb capacity,<sup>9,49,50</sup> although this test has not yet been validated for children with unilateral CP.

There is still no golden standard to assess the amount of upper limb use, which is considered the main parameter of developmental disregard. In this thesis we developed (or revised) two upper-limb measures to assess this specific aspect of upper limb performance and provided a first indication of their psychometric properties. Many studies<sup>6, 51, 52</sup> have used the *frequency* scores from the "how-often" scale of the Pediatric Motor Activity Log (PMAL<sup>47</sup>), whereas some studies have used the Shriners Hospital Upper Extremity Evaluation,<sup>53</sup> the Observational Skills Assessment Score<sup>54</sup> or the Assisting Hand Assessment<sup>55</sup> as a measure for amount of use. The notion that typically developing children use both hands fluently and almost constantly during bimanual activities while children with unilateral CP often use their affected side only when they are forced to, constituted the basis for developing and revising the VOAA-DDD(-R) (chapter 6). The VOAA-DDD-R took a novel approach to establish developmental disregard based on a cut-off criterion for the difference in *duration* of use between two specifically designed tasks: one task *demanding* and the other task merely *provoking* a bimanual task approach. The test was revised and validated for children between 2.5 and 8 years. The validation of an adapted version for children in the age range from 8 to 12 years was recently completed.<sup>56</sup> This adapted version requires older children to perform the activities as quickly as possible, appealing to the automatic use of their affected arm and hand, which improved the sensitivity to identify developmental disregard in these older children.

In chapter 7 we argued that, in addition to clinical testing, a parent-rated questionnaire was needed to assess the spontaneous use of the affected upper limb of children with unilateral paresis in the home environment, as the primary goal of upper limb interventions is the enhancement of automatic use of the affected side in daily activities. Moreover, by asking parents how they perceive the spontaneous hand use of their child at home we acknowledge their expertise in this matter. At the same time, it provides a good opportunity for a dialogue between therapists and parents about their observations and expectations before an intervention. In chapter 7 we described how the Hand-Use at Home questionnaire (HUH) was developed and tested for its unidimensionality, hierarchical properties and construct validity in collaboration with parents of children with unilateral paresis. A subsequent study is currently under way reporting other psychometric properties of the HUH<sup>57</sup> including additional construct validity, good test-retest reliability (ICC = 0.89), and good absolute agreement. As we hypothesize that children with developmental disregard need more time to automatize the use of their affected hand in bimanual activities, an important next step will be to establish whether this lack of automatization is reflected in a difference of HUH-scores between children with and those without developmental disregard.

## Recommendations for clinical practice and research

The Dutch health-care system is going through major changes to control the increasing costs of care and to sustain and improve health-care delivery for the future. Service

providers are challenged to review their 'best care' with respect to the changing society, cost-effectiveness and delivering individually tailored services. For patients and people with chronic diseases the concept of 'participation' is extended to taking responsibility for their own health and care and becoming a partner in clinical decision making. As for children with unilateral CP, the effective use of the hand in age-appropriate and daily activities is an important point of interest throughout their childhood and will often require more than just one period of upper limb training. Children with a chronic condition such as CP often need clinical support to be able to adapt to their (changing) environment as they experience physical and cognitive/mental challenges more than typically developing children in their struggle for independency and becoming an adult. With the changing health-care system the cost-effectiveness of intensive group interventions delivered from rehabilitation centers as described in this thesis has to be considered critically. CIMT-BiT interventions carried out in more natural environments (e.g. home, school) may be less costly and the effectiveness of home-based training may be similar to clinical intervention programs. An important prerequisite, however, is that family members and teachers carrying out or supporting the training program need to be properly trained and supervised<sup>1</sup>. A disadvantage of home-based interventions with parents combining the roles of educator and trainer is the risk of unacceptable parental stress and deterioration of the normal parent-child interactions, which may result in decreasing therapy adherence. Therefore, home-based interventions require a collaborative relationship between therapists and families to balance the needs of the children with the capacities of the families and to provide adequate support. The use of home-based training as an addition to (or as an alternative for) clinical interventions may be a suitable way to enhance the continuity of upper limb stimulation and practice in daily life, involving parents and significant others. Hence, service providers should devote efforts to develop and implement efficient and innovative ways to support and empower children and their families to manage the consequences of neurological impairments and to attain functional independence and societal participation.

The studies in this thesis have provided evidence for the effectiveness of upper limb CIMT-BiT interventions as well as for the validity of three new outcome measures to assess improvements of upper limb capacity and performance in (young) children with unilateral CP. The findings of these studies and the reflections in this discussion lead to the following clinical and scientific recommendations.

***Recommendations to improve upper limb interventions in children with unilateral CP:***

- The choice for a CIMT, BiT or CIMT-BiT approach should be based on what fits most closely to the characteristics, age and preferences of the individual child (and preferences of its parents).
- Children from the age of 5 years should be involved in the process of goal setting.

- Parents and children need to be integral members of the treatment team such that individualized services and tools for age-appropriate practice at home can be offered.
- Development of a self-management (group) intervention for adolescents with unilateral CP addressing general problem-solving skills to deal with current and future challenges in a broader perspective than with the mere aim to promote functional use of the affected upper limb should be considered.
- Studies should be conducted to examine the feasibility and effectiveness of home-based training interventions to reduce developmental disregard and improve automatic use of the affected upper limb in children with unilateral CP.
- During home-based intervention programs, parental stress should be carefully monitored.
- Future studies are warranted to find evidence whether an approach aimed at reduction of cognitive load during bimanual activity reduces developmental disregard in children with unilateral CP.
- Future research should assess the feasibility and effectiveness of a higher dosage of CIMT-BiT training (currently 72 hours) in children aged 5 to 8 years to promote the stabilization of learned skills.

***Recommendations to improve upper limb outcome assessments in children with unilateral paresis:***

- Clinicians should only use tests and questionnaires with sound psychometric properties to assess outcomes of an intervention.
- Through international consensus, a core set of instruments assessing the most important aspects of upper limb functions, capacities and performance should be developed to enhance comparability between studies in children with unilateral CP.
- Future research should investigate the responsiveness to change of the VOAA-DDD-R and the HUH, and examine the relationship between these measures of amount of affected hand use.
- New studies need to establish whether the lack of automaticity of affected hand use is reflected in significantly different HUH scores between CP children with and without developmental disregard.
- Future research should investigate whether the development of a HUH self-rating questionnaire for CP children between 11 and 18 years would be feasible to assess daily life hand use.
- Future studies should apply brain-imaging techniques (e.g. electroencephalography / EEG, transcranial magnetic stimulation / TMS, magnetic resonance imaging / MRI) to investigate the effects of intensive upper limb intervention on neuroplasticity in children with unilateral CP.

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# 9

Nederlandse samenvatting



Met een prevalentie van 2 tot 2,5 per 1000 levend geboren kinderen is cerebrale parese (CP) een van de meest voorkomende oorzaken van motorische beperkingen bij kinderen. Ongeveer 30% van deze kinderen wordt geclassificeerd als unilaterale spastische CP. Dit betekent dat bij deze kinderen de motorische beperkingen vooral aan de linker of rechter zijde van het lichaam optreden. In de meeste gevallen zijn de beperkingen van de bovenste extremiteit groter dan van de onderste extremiteit. Het effectieve gebruik van de arm en hand voor het reiken, grijpen, vasthouden, loslaten of manipuleren van voorwerpen is lastig en belemmert het uitvoeren van dagelijkse tweehandige activiteiten. Zelfs als de motorische vaardigheid van de aangedane arm en hand vrij goed is kan er bij kinderen met unilaterale CP een duidelijke discrepantie bestaan tussen hun 'capacity'<sup>3</sup> (mogelijkheden om de hand te gebruiken onder optimale condities) en 'performance'<sup>1</sup> (hoe de hand daadwerkelijk wordt gebruikt in het dagelijks leven). Deze kinderen gebruiken hun arm en hand in het dagelijks leven duidelijk minder dan dat op basis van hun mogelijkheden verwacht mag worden. Dit wordt ook wel 'developmental disregard' (DD) genoemd.

Gebaseerd op experimenten met apen en op het onderzoek naar effectieve interventies bij volwassenen na een hersenbloeding zijn er nieuwe behandelprotocollen ontwikkeld voor kinderen met unilaterale CP. Deze zijn gericht op het verbeteren van het daadwerkelijk dagelijks gebruik van de aangedane arm en hand. Zowel 'constraint induced movement therapy' (CIMT) als bimanuele training (BiT) zijn specifieke behandelingen die bestaan uit een korte intensieve periode van specifieke training van de arm en hand. Het is de bedoeling om daarmee de mogelijkheden voor het gebruik van de aangedane arm en hand te vergroten, het gecoördineerd samenwerken van beide handen te verbeteren en DD te verminderen. Het uiteindelijke doel is het verbeteren van de uitvoering van dagelijkse activiteiten. Het meest opvallende verschil tussen CIMT en BiT is dat bij de CIMT gebruik gemaakt wordt van een sling of spalk om de niet-aangedane hand te beperken, terwijl de aangedane arm en hand worden getraind. Bij BiT worden de arm en hand intensief getraind door middel van tweehandige activiteiten. Om de uitvoerbaarheid en effectiviteit van training voor jonge kinderen (2,5 tot 8 jaar) te vergroten hebben we het originele CIMT-protocol aangepast tot een kindvriendelijke interventie. Deze 'modified CIMT' (mCIMT) wordt gecombineerd met een aanvullende periode BiT (mCIMT-BiT) om daarmee de vaardigheidswinst van de aangedane arm en hand te integreren in tweehandige activiteiten die passen bij de leeftijd en de beleevingswereld van het kind. Voor oudere kinderen met unilaterale CP (8 tot 18 jaar) is dit mCIMT-BiT protocol verder aangepast en gecombineerd met zelfmanagement training gedurende de bimanuele trainingsfase. Door deze toevoeging proberen we oudere kinderen te stimuleren om het efficiënte gebruik van hun aangedane hand bewust en zelfstandig te monitoren zonder

3 Terminologie volgens internationale classificatie voor functioneren, beperkingen en gezondheid bij kinderen en jongeren (ICF-CY)

dat zij hierbij door volwassenen hoeven te worden aangespoord. Het gecombineerd gebruik van CIMT en BiT in één interventie was een nieuwe benadering in de behandeling van kinderen met unilaterale CP. De effectiviteit van deze combinatietherapie was niet eerder getest. Ook de combinatie van mCIMT-BiT met zelfmanagement training was nieuw en nog niet eerder onderzocht.

Hoewel de effectiviteit van intensieve behandelingen voor de arm en hand wordt gemeten met behulp van valide en betrouwbare uitkomstmaten, is er een aantal functionele aspecten van zowel 'capacity' als 'performance' dat niet voldoende specifiek kan worden gemeten, omdat in de bestaande testen voldoende detaillering ontbreekt. Zo kan bij jonge kinderen (2,5 tot 8 jaar) de capaciteit voor grijpen, vasthouden en loslaten niet goed onderzocht worden met de huidige 'capacity' testen voor deze doelgroep, omdat deze te weinig object-gerelateerde items bevatten. Andere testen meten weliswaar de vaardigheid bij grijpen, vasthouden en loslaten, maar vereisen dat de handelingen zo snel mogelijk worden uitgevoerd en zijn diengevolge niet geschikt voor jonge kinderen. Daarom hebben we onderzocht of de recent gepubliceerde Modified House Classification (MHC), die de vaardigheden grijpen, vasthouden en loslaten onderzoekt door middel van 32 object-gerelateerde items, wél geschikt was voor kinderen met unilaterale CP.

Met betrekking tot het meten van 'performance' vonden we onvoldoende valide en betrouwbare instrumenten om te meten hoe vaak de aangedane arm en hand spontaan worden gebruikt door kinderen met unilaterale CP. Dat leidde tot verfijning en revisie van de Video Observatie Aarts en Aarts-Determine Developmental Disregard (VOAA-DDD). Deze test meet 'capacity', 'performance', en hoe veel de hand wordt gebruikt bij een tweetal tweehandige taken. De validiteit en test-hertest betrouwbaarheid van de gereviseerde VOAA-DDD (VOAA-DDD-R) zijn onderzocht. Klinische testen zijn echter niet geschikt en ook niet ontworpen om het daadwerkelijke spontane gebruik van de hand te meten in dagelijkse situaties en natuurlijke omstandigheden. Derhalve is een nieuwe ouder vragenlijst ontwikkeld om het spontane dagelijkse handgebruik meetbaar te maken en zijn de klinimetrische eigenschappen van deze test onderzocht.

Dit proefschrift begint met een algemene inleiding (hoofdstuk 1) waarin achtergrondinformatie wordt gegeven over unilaterale CP, over intensieve behandelingen voor de arm en hand, en over het meten van 'capacity', 'performance', en de mate van gebruik van arm en hand bij deze groep kinderen. Het vervolg van het proefschrift bestaat uit twee delen.

In deel 1 ligt het accent op de behandeling van de arm en hand bij kinderen met unilaterale CP. Het start met twee studies die de effectiviteit bestuderen van de kindvriendelijke mCIMT-BiT interventie voor jonge kinderen (de Piratengroep). Daarnaast wordt een haalbaarheidsonderzoek beschreven van de CIMT-BiT interventie in combinatie met zelfmanagement training, welke voor oudere kinderen met unilaterale CP is ontwikkeld. Deel 2 behandelt de functionele beoordeling van de arm en hand bij kinderen met

unilaterale CP. In dit deel wordt de revisie beschreven van twee observatieve testen (de MHC en de VOAA-DDD-R) en wordt het onderzoek naar de klinimetrische eigenschappen van deze testen beschreven. De derde studie in deel twee beschrijft de ontwikkeling van de Hand-Use-at Home vragenlijst (HUH) en het eerste onderzoek naar de interne structuur, unidimensionaliteit en validiteit hiervan.

## **Deel een: Interventies voor de arm en hand bij unilaterale CP**

### **Hoofdstuk 2**

In dit hoofdstuk rapporteren we het onderzoek naar de effectiviteit van een nieuwe, speelse groepsbehandeling voor jonge kinderen met unilaterale CP. Deze mCIMT-BiT interventie (ook wel Piratengroep genoemd) begon met 6 weken mCIMT en werd gevolgd door 2 weken BiT. Door middel van een gerandomiseerd onderzoek met controlegroep (RCT) is onderzocht of deze mCIMT-BiT interventie het spontaan gebruik van de aangedane arm en hand verbeterde, zowel in kwalitatief als kwantitatief opzicht, in vergelijking met reguliere behandeling van eenzelfde duur. Voor dit onderzoek werden kinderen gerekruteerd tussen de 2,5 en 8 jaar oud met unilaterale spastische CP en een manual ability classification system (MACS) score van I, II of III. Zij werden gerandomiseerd voor de mCIMT-BiT interventie of voor de controlegroep die reguliere behandeling kreeg.

De mCIMT-BiT groep (n=28) kreeg wekelijks gedurende 3 middagen een training van 3 uur (9 uur per week). In de eerste 6 weken werd er totaal 54 uur mCIMT gegeven, gevolgd door 2 weken taak-specifieke training tijdens doelgerichte bimanuele spel- en zelfverzorgingsactiviteiten (18 uur BiT). De reguliere behandeling van de controlegroep bestond uit 1,5 uur fysiotherapie en/of ergotherapie per week, aangevuld met dagelijkse oefeningen (minimaal 7,5 uur per week, met ouders en/of leerkracht). De oefeningen waren gericht op het optimaal gebruik van de aangedane arm en hand. Primaire uitkomstmaten waren de Assisting Hand Assessment (AHA) en de ABILHAND-Kids. Secondaire uitkomstmaten waren de Melbourne assessment of Unilateral Limb Function (Melbourne), de Canadian Occupational Performance Measure (COPM), en de Goal Attainment Scale (GAS). Alle primaire en secundaire uitkomstmaten (uitgezonderd de Melbourne) lieten significante verbeteringen zien in de mCIMT-BiT groep. Deze verbeteringen bleven behouden bij de tweede nameting (8 weken na de behandeling). In de controlegroep daarentegen werden geen significante veranderingen gevonden. We concludeerden dat mCIMT gevolgd door taakspecifieke training tijdens doelgerichte bimanuele spel- en zelfverzorgingsactiviteiten een effectieve interventie is om het spontaan gebruik van de aangedane arm en hand te verbeteren bij kinderen met unilaterale CP die een relatief goede arm-hand functie hebben bij aanvang van de behandeling.



### Hoofdstuk 3

Alle kinderen die deelnamen aan de RCT (beschreven in hoofdstuk 2) werden 6 maanden na het einde van de behandeling nogmaals getest om de lange termijn resultaten van de interventie te bepalen. De primaire (AHA, ABILHAND-Kids) en secundaire (Melbourne, COPM) uitkomsten van beide groepen werden vergeleken zowel tussen de groepen als binnen iedere groep met de resultaten van de eerste nameting (1 week post interventie). Aanvullend werden de kinderen van de mCIMT-BiT groep 1 jaar na de interventie voor een laatste maal getest; vijf van hen waren niet meer beschikbaar voor deze laatste nameting.

De primaire uitkomstmaten na 6 maanden bleken stabiel in de tijd zowel in de interventiegroep als in de controlegroep. In de mCIMT-BiT groep bleven de resultaten zelfs stabiel tot een jaar na de interventie. Ook de scores voor uitvoering en tevredenheid van de vijf belangrijkste doelen die gemeten werden met behulp van de COPM bleken 6 maanden na de interventie stabiel in beide groepen. De unilaterale capaciteit (gemeten met de Melbourne) liet in de periode tussen 1 week en 6 maanden na de interventie een significante verbetering zien in de mCIMT-BiT groep, maar niet in de controlegroep. De bevindingen waren coherent met eerdere studies die de resultaten van CIMT en BiT interventies met elkaar vergeleken. In onze studie bleek dat 72 uur intensieve mCIMT-BiT eenzelfde behoud van resultaat liet zien als CIMT of BiT interventies van 90 uur.

In een tweede studie werd ook de verandering in handvaardigheid gemeten bij kinderen in de mCIMT-BiT groep om het maximale trainingseffect van de interventie vast te stellen gedurende de 6 weken mCIMT (54 uur) en de daarop volgende twee weken BiT (18 uur). Het doel was het verkrijgen van extra informatie die kan helpen bij het vaststellen van de optimale dosering van intensieve training van de bovenste extremiteit bij jonge kinderen met unilaterale CP. Bij alle kinderen van de interventiegroep ( $n=28$ ) werd wekelijks de Box and Blocks test (BBT) afgenomen. Het aantal blokjes dat gedurende 1 minuut werd verplaatst met de aangedane hand werd hierbij genoteerd en de ruwe score werd gebruikt om de individuele leercurves vast te stellen. Twee geblindeerde onderzoekers stelden voor ieder kind vast waar de top van de leercurve (maximaal individueel trainingseffect) werd bereikt gedurende de interventie. Vijftien kinderen (53,6 %) bereikten een maximaal trainingseffect binnen de 6 weken mCIMT. Deze groep kinderen verschilde van de overige kinderen met betrekking tot leeftijd, maar niet wat betreft geslacht, aangedane hand of MACS score. Kinderen onder de 5 jaar hadden een grotere kans om een maximale BBT score te bereiken binnen de eerste 6 weken mCIMT dan oudere kinderen. Dit maximum stabiliseerde vaak al na 4 weken unimanuele training. Oudere kinderen lieten gedurende een langere periode vooruitgang zien in hun handvaardigheid, terwijl hun scores veelal lager werden gedurende de periode van bimanuele training. Deze bevindingen suggereren dat kinderen van 5 jaar en ouder mogelijk kunnen profiteren van een langere periode unimanuele training dan 54 uur om hun maximale handvaardigheid te bereiken, te stabiliseren en te behouden gedurende de periode van bimanueel trainen.

## Hoofdstuk 4

Intensieve training van de arm en hand bij oudere kinderen en adolescenten (8-18 jaar) met unilaterale CP is over het algemeen alleen mogelijk als deze aangeboden wordt in de vorm van een trainingskamp gedurende schoolvakanties. Tot nu toe werden in de literatuur alleen kampen beschreven met ofwel een CIMIT ofwel een BiT interventie. In deze studies werd 30 tot 90 uur training aangeboden tijdens een periode van 1 tot 3 weken. De kritische dosis CIMIT of BiT die noodzakelijk is om betekenisvolle veranderingen teweeg te brengen in unimanuele capaciteit en tweehandige uitvoering van activiteiten is nog steeds onderwerp van discussie. Derhalve hebben we onderzocht of een gecombineerde CIMIT-BiT interventie van 1 week haalbaar en voldoende effectief is om zowel de unilaterale capaciteit van arm en hand als ook de uitvoering van tweehandige activiteiten te verbeteren bij kinderen met unilaterale CP tussen 8 en 18 jaar oud. Bij deze interventie waren de individuele doelen leidend. Zelfmanagement training werd geïntegreerd in de CIMIT-BiT interventie om het effect te maximaliseren en de jongeren in staat te stellen het effectieve gebruik van hun aangedane arm en hand zelfstandig te monitoren.

Functionele doelen (COPM), unimanuele 'capacity' (Box and Blocks test), bimanuele vaardigheden (ABILHAND-Kids en Children's Hand-use Experience Questionnaire / CHEQ), en de mate van inschakelen van de hand (VOAA-DDD-R) werden gemeten voorafgaand aan de start van de behandeling (baseline) evenals 1 week en 4 maanden na de behandeling. Twintig kinderen (gemiddelde leeftijd 9,5 jaar) namen deel aan het onderzoek. Bij de nametingen vonden we significante verschillen ten opzichte van de baselinemeting op alle uitkomstmaten. De grootste effecten vonden we voor de COPM-scores voor uitvoering en tevredenheid van de individueel vastgestelde doelen (respectievelijk Cohens  $d=2,09$  en  $d=2,42$ ). Het effect was groot voor de ABILHAND-Kids ( $d=0,86$ ), gemiddeld voor de CHEQ ( $d=0,70$ ) en Box en Blocks test ( $d=0,56$ ), en klein voor de VOAA-DDD-R ( $d=0,33$ ). Alle resultaten bleven behouden bij de meting na 4 maanden. De resultaten van deze studie laten zien dat één week (36 uur) van intensieve mCIMIT-BiT gecombineerd met zelfmanagement training haalbaar is voor oudere kinderen met unilaterale CP en veelbelovend is om arm-hand vaardigheden en tweehandige activiteiten te bevorderen.

## Deel twee: Functionele beoordeling van de arm en hand bij unilaterale CP

### Hoofdstuk 5

Veel kinderen met unilaterale CP hebben beperkte vaardigheden om voorwerpen te hanteren met de aangedane hand. Dit hindert hen bij het uitvoeren van tweehandige activiteiten. Er zijn diverse meetinstrumenten om de capaciteit van de arm en hand te meten bij kinderen met unilaterale CP. Het ontbreekt echter aan een valide instrument dat specifiek de vaardigheden grijpen, vasthouden en loslaten bij tweehandige activiteiten meet. De MHC is door Koman et al.<sup>4</sup> geïntroduceerd met daarin 32 items die gerelateerd zijn aan het functioneel gebruik van de aangedane arm en hand. De somscore (aantal behaalde items) representeert de functionele capaciteit van de onderzochte hand (range 0 – 32 punten). In deze studie hebben we onderzocht of de MHC gebruikt kan worden om een objectief onderscheid te maken tussen verschillende niveaus van functionele capaciteit van de aangedane arm en hand bij kinderen met unilaterale CP. We hebben de unidimensionaliteit en item-hiërarchie geëvalueerd in een groot cohort van kinderen met unilaterale CP (kalibratiecohort). De MHC items zijn gescoord van 369 assessments die bij 159 kinderen met een unilaterale CP (gemiddelde leeftijd van 6,5 jaar, range 2,1 – 17,4 jaar) waren afgenomen en op video opgenomen. We gebruikten een Rasch-analyse om items te elimineren, te ordenen en te wegen en hiermee de klinimetrische eigenschappen van het instrument te verbeteren. De geëxcludeerde items waren te gemakkelijk of te moeilijk voor de kinderen in onze steekproef. Veertien items bleken samen een goed construct te vormen om functionele capaciteit te meten. Constructvaliditeit van deze item-set is vervolgens getest binnen het kalibratiecohort als ook in een nieuwe steekproef van 40 kinderen met unilaterale CP en een gemiddelde leeftijd van 8,1 jaar (validatiecohort). Het bleek dat de gewogen somscore van de 14-item MHC sterk correleerde met de MACS-classificatie en een vrij goede overeenkomst vertoonde met de ABILHAND-Kids.

We concludeerden dat de originele volgorde van items van de MHC verbeterd kan worden en de item-set ingekort, zodat de somscore gebruikt kan worden voor het meten van kinderen met unilaterale CP. De Rasch-gereduceerde 14-item set met gewogen somscore toont een goede constructvaliditeit om de functionele capaciteit van de aangedane arm en hand te meten bij kinderen met unilaterale CP.

### Hoofdstuk 6

In deze studie werd de validiteit en betrouwbaarheid van de VOAA-DDD-R geëvalueerd. Deze test is gericht op het meten van 'capacity', 'performance' en 'developmental disregard' van de aangedane arm en hand bij kinderen met unilaterale CP. De test bestaat

4 Koman LA, Williams RM, Evans PJ, Richardson R, Naughton MJ, Passmore L, et al. Quantification of upper extremity function and range of motion in children with cerebral palsy. *Dev Med Child Neurol*. 2008; 50: 910-7

uit twee gestandaardiseerde tweehandige taken die opgenomen worden op video en off-line worden gescoord. De eerste taak lokt het gebruik van beide handen uit (muffin versieren), terwijl de tweede taak het gebruik van beide handen vereist (kralen rijgen). In deze gereviseerde versie van de VOAA-DDD werd het scoringssysteem vereenvoudigd en werden de taken zodanig aangepast dat zowel het contrast tussen beide taken als de overeenkomst in observeerbare handelingen van de aangedane hand werden vergroot. In beide taken werd de frequentie van grijpen, vasthouden en loslaten gemeten evenals de duur van het gebruik van de aangedane hand. De test genereert 3 scores. De frequentiescore van de rijgtaak wordt gebruikt om de 'capacity' (0-100 %) te berekenen en met de frequentiescore van de muffintaak wordt de 'performance' score (0-100 %) berekend. Het verschil tussen de duurscores van beide taken wordt gebruikt als maat voor developmental disregard (0-100 %).

De VOAA-DDD-R werd twee keer afgenomen bij vijftientig kinderen met unilaterale CP (tussen 2,5 en 8 jaar oud) en bij 46 normaal ontwikkelende kinderen van vergelijkbare leeftijd en geslacht. Constructvaliditeit werd bepaald door een vergelijking tussen de scores van kinderen met unilaterale CP en die van normaal ontwikkelende kinderen. Interbeoordelaar-, intrabeoordelaar-, en test-hertest betrouwbaarheid werden bepaald met behulp van de intraclass correlatie coëfficiënt (ICC). Tevens werden de standaard meetfout (SEM) en het kleinste meetbare verschil (SDD) berekend. Voor het identificeren van developmental disregard werd een afkapwaarde berekend die werd vastgesteld op basis van de gemiddelde score van normaal ontwikkelende kinderen (+ 2 standaarddeviaties).

Uit de resultaten bleek dat kinderen met unilaterale CP significant lager ( $p < 0,05$ ) scoorden dan de normaal ontwikkelende kinderen op 'capacity' (77% versus 98%) en 'performance' (55% versus 100%) en veel hogere scores ( $p < 0,05$ ) hadden voor 'developmental disregard' (23% versus 7%). De ICC's waren hoog (0,79 – 1,0) en duiden op goede betrouwbaarheid. De SEM's en SDD's varieerden tussen respectievelijk 4,5% – 6,8% en 12,5% – 19,0%. Concluderend zijn de drie scores van de VOAA-DDD-R ('capacity', 'performance' en 'developmental disregard') valide en betrouwbaar te gebruiken om de specifieke arm-handproblemen bij kinderen met unilaterale CP in kaart te brengen. De resultaten bevestigen dat veel kinderen met unilaterale CP een discrepantie laten zien tussen gebruik van de aangedane hand bij een taak die tweehandig werken vereist ('capacity') en een taak die tweehandig werken slechts uitlokt ('performance'). De VOAA-DDD-R scores kunnen gebruikt worden om deze specifieke problemen bij het gebruik van de aangedane arm en hand bij kinderen met unilaterale CP te objectiveren, zodat interventies specifiekere kunnen worden ingezet.

## Hoofdstuk 7

Bij kinderen met unilaterale CP of een obstetrische plexus brachialis laesie (NBPP) kan het herhaaldelijk falen van pogingen om activiteiten uit te voeren met de aangedane arm en

hand leiden tot een verminderd spontaan gebruik van de hand tijdens dagelijkse activiteiten. Het gevolg is dat kinderen met een unilaterale parese hun aangedane hand minder vaak gebruiken bij tweehandige activiteiten dan verwacht zou mogen worden op basis van hun functionele capaciteiten. Dit wordt ook wel aangeduid met de term 'developmental disregard'. Ouders geven vaak aan dat hun kind, ondanks intensieve revalidatie, de aangedane arm en hand minder gebruikt dan mogelijk is en veel activiteiten éénhandig uitvoert. Ondanks het feit dat het fenomeen 'developmental disregard' als een belangrijk probleem wordt gezien, bestaat er nog geen valide en betrouwbaar meetinstrument om het spontane gebruik van de aangedane arm en hand tijdens het dagelijks leven te meten. Een vragenlijst voor ouders die het spontane gebruik van de aangedane hand tijdens dagelijkse activiteiten meet zou een waardevolle aanvulling zijn op de bestaande testen en vragenlijsten. In dit hoofdstuk wordt de ontwikkeling van de Hand-Use at Home (HUH) vragenlijst als ook het onderzoek naar de interne structuur en constructvaliditeit hiervan beschreven. De HUH evalueert hoe vaak de aangedane arm en hand worden gebruikt tijdens dagelijkse activiteiten die zeer regelmatig voorkomen bij jonge kinderen van 3 tot 10 jaar oud. Ouders scoren bij elke activiteit het spontane gebruik van de aangedane hand van hun kind op een schaal met 5 antwoordcategorieën. De totaal score van de HUH geeft de mate weer waarin het kind zijn aangedane hand in de thuissituatie spontaan inzet. Om de validiteit van de somscore te testen zijn de structuur van de antwoordschaal en de unidimensionaliteit en item-hiërarchie onderzocht met behulp van Rasch-analyse. Data van 322 kinderen (gemiddelde leeftijd 6,6 jaar, SD 2,1 jaar) met unilaterale CP ( $n=131$ ) of NBPP ( $n=191$ ) werden verzameld. Daarnaast werden ook HUH vragenlijsten van 55 normaal ontwikkelende kinderen (TD) (6,8 jaar, SD 2,5 jaar) gebruikt om de constructvaliditeit vast te stellen. Rasch-analyse toonde aan dat de antwoordschaal met 5 categorieën niet rechtstreeks gebruikt kon worden voor berekening van een somscore; omzetting naar een 3-puntscore gaf een goede antwoordstructuur. Het onderzoek liet zien dat 18 hiërarchisch geordende activiteiten, gescoord op een schaal met 5 antwoordcategorieën en omgezet naar een 3-puntscore, een passend Rasch-model vormen. De goede spreiding van de geordende items laat zien dat deze spontaan gebruik meten van 'gemakkelijk' naar 'moeilijk'. De HUH somscore liep van  $-4,69$  tot  $+5,17$  logits. Interne consistentie van de HUH was hoog (Cronbach's  $\alpha = 0,941$ ).

De HUH-score was in staat te discrimineren tussen kinderen met unilaterale CP, kinderen met NBPP en normaal ontwikkelende kinderen ( $p < 0,001$ ) en bevestigde zo de constructvaliditeit van de HUH en de opgestelde hypothesen. De groep normaal ontwikkelende kinderen toonde de hoogste mate van spontaan gebruik van de arm en hand (mediaan 2,51 logits) met 5,5 % kinderen die een maximale score lieten zien. De laagste HUH score werd, zoals verwacht, gevonden bij de CP groep (mediaan  $-0,343$  logits) en de NBPP groep scoorde tussen beide andere groepen met een mediane HUH-score van  $+1,07$  logits. De HUH blijkt een valide uitkomstmaat om het spontane gebruik van de aangedane arm en hand te meten bij kinderen met een unilaterale parese

en een leeftijd van 3 tot 10 jaar oud. Aanvullende studies zijn nodig om de test-hertest betrouwbaarheid en de sensitiviteit voor verandering van de HUH te onderzoeken.



# 10

Dankwoord  
About the author





## Dankwoord

Hier ligt dan mijn druppel op de gloeiende plaat van de wetenschap!

Dit boekje is het product van een aantal jaren onderzoeken, lezen, leren, denken, ontdekken, discussiëren, luisteren, schrijven, wachten, mopperen en blijdschap. Hoewel ik promoveren ook wel als een eenzame exercitie heb ervaren is het vooral een traject geweest voor mensen en met mensen. Er is een groot aantal mensen geweest dat heeft meegeleefd, meegedacht, ondersteund en geholpen. Deze laatste pagina's van mijn boekje zijn voor hen.

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Mijn ouders hebben aan de basis gestaan van mijn ontwikkeling en hebben van jongs af aan mijn 'waarom' en 'hoe' altijd gestimuleerd met "Zoek maar op!" De woordenboeken en encyclopedie in de boekenkast thuis hebben zeker niet staan verstoffen. Dankjewel mam, en ik denk dat papa best wel trots zou zijn.

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De interventies die beschreven zijn in dit boekje zouden niet zo succesvol zijn geweest zonder mijn collega's van de kinderrevalidatie die met enorme passie en expertise handen en voeten hebben gegeven aan de organisatie en behandelingen in de Piratengroep en de Hou van Hollandgroep.

Het werken met de kinderen en hun ouders is ontzettend inspirerend en zij zijn ook degenen voor wie deze onderzoeken zijn gedaan. Beste ouders, wat heb ik een bewondering voor jullie doorzettingsvermogen bij de zoektocht naar het beste voor jullie kinderen. Dank voor de openheid bij onze gesprekken over wensen en doelen, het vertrouwen dat jullie ons gaven en de ruime bereidheid om mee te doen aan de onderzoeken.

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## About the author

Yvonne Geerdink was born on July 29, 1959 in Oss, the Netherlands. After finishing secondary education (VWO) at the Titus Brandsma Lyceum in Oss, she studied occupational therapy (OT, Bachelor level) at the OT school for Higher Education in Hoensbroek from 1978 to 1982. Yvonne started her professional life as an OT in a nursing home with a large group of adolescent and young adult residents with physical disabilities and with a pediatric department with very young children as well. She started specializing in pediatric occupational therapy. Her main expertise was the development of hand function and writing skills in children with learning disabilities. Throughout her career, Yvonne worked in the field of pediatric occupational therapy in settings that vary from a medical day-care center for children with severe and multiple disabilities, to a special school for children with epilepsy. Simultaneously she ran a private occupational therapy practice for five years in Boxtel. Since 1997, she works as an occupational therapist in pediatric rehabilitation.

To better understand statistical analysis of scientific papers on pediatric interventions and to gain knowledge of the psychometric properties of tests, she followed the Master Evidence Based Practice at the University of Amsterdam-Amsterdam Medical Center. In 2010 she graduated as a clinical epidemiologist (MSc). She was involved in the development of de VOAA–DDD(-R) and the effect study on the Pirate group intervention as assessor and co-author. In 2011 Yvonne started her PhD research at the Sint Maartenskliniek in Nijmegen on refined assessment instruments for the arm and hand and the (long-term) effectiveness of upper limb interventions (mCIMT-BiT) for children with unilateral Cerebral Palsy.

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